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CONGENITAL IMPERFORATE RECTUM, RECTO-URETHRAL AND RECTO-VAGINAL FISTULAE

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THE purpose of this paper is to describe the *levator ani* musculature in relation to congenital imperforate rectum, and to discuss its surgical application in reconstruction of the anal canal in this deformity.

Wood Jones (1904) distinguished between anal and rectal deformities. Abnormalities of development of the proctodeal pit and membrane constitute the anal group, which is more common and more easily treated (Denis Browne, 1951) and which provides the greatest number of satisfactory surgical cures. The rectal group consists of those abnormalities in which the terminal end of the hind-gut ends at the level of the *verumontanum* or higher in the male, and in the female lies at a corresponding level in the pelvis or opens into the posterior wall of the vagina. These are referred to as imperforate rectum, with or without a communicating fistula to the urinary tract or vagina. It is this group of visceral deformities, together with the associated deformities of the spine, sphincters and *levator ani*, with which this paper is concerned.

Embryological Subdivisions of the Rectum

The rectum and anal canal develop from the hind-gut, the cloaca and the proctodeal pit. The line of the anal valves delimits the proctodeal segment. The rectum above this to its origin at the third sacral vertebra is derived from the cloaca and hind-gut. The embryological dividing line between the cloacal rectum and that formed directly from hind-gut is not certain. Wood Jones (1911) described the hind-gut contribution as ending at the level of the peritoneal reflection or Houston's third fold, where anatomical changes in the bowel suggest a different embryological origin. Though it is difficult to reconcile his "post-allantoic gut" theory of development of the rectum with later studies which support the theory of subdivision of the cloaca, the congenital abnormalities described in this paper fall into groups which conform to Wood Jones' embryological rectal boundaries.

Common Deformities

The common deformities can be correlated with abnormal formation of these rectal segments.

(A) Anal deformities

These occur below the level of the valves and concern the proctodeal segment.

1. Ectopic anus. — This is a well-formed proctodeal pit in an anterior position.
2. Imperforate anus or anal stenosis. This is due to complete or partial persistence of the proctodeal membrane. This occurs in a normally situated or ectopic anus and the membrane is deeply placed approximately at the level of the triangular ligament in the male. This is a rare condition in the female and no case was studied in this series.

3. "Covered" anus (Denis Browne, 1951). The anus is formed either in the normal or ectopic position and is covered in completely or partially by abnormal posterior fusion of the inner genital folds (Wood Jones, 1915). The perineal *raphe* is extended posteriorly over the site of the anus and is usually thickened or raised to form a tag. The skin on either side of the *raphe*, or in front of or behind the tag is thin, transparent or perforated. The membrane of this type is superficial on the plane of the perineum.

(Photo by Fayer)

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The fusion of the folds takes place from behind forwards, and sometimes the attenuated anal lumen is rolled forwards to open at some point in the perineum, scrotum or penis in the line of the *raphe*. Cases have been reported where the track leads to the region of the *frenulum*.

The lumen of the anus may be projected anteriorly at a deeper level into the bulb of the urethra, in which case an associated atypical hypospadiac deformity may coexist. This is an ano-bulbar fistula and is not to be confused with recto-urethral fistula.

In the female, the anus may be projected forwards into the perineum by abnormal fusion of the inner genital folds, most commonly to open at the region of the fourchette. This must not be confused with recto-vestibular fistula. Frequently a *raphe* can be seen posterior to the fistula traversing the perineum. More exaggerated fusion of the folds occludes the vestibule as well as the anus, which is then projected forwards, together with the vagina and urethra, to the base of the clitoris. These types are described for completeness of the classification. Their management is discussed by Denis Browne (1951).

(B) Rectal deformities

In this investigation 16 males and 13 females with rectal deformities were studied. These concern the middle rectal segment. Due to failure of subdivision of the cloaca, the terminal end of the hind-gut contribution to the rectum lies at or about the level of the third fold of Houston, attached by fibrous tissue or a fistulous track, to the undivided cloaca or to the vagina.

In the male, the rectum may end blindly, remaining attached to the cloaca by fibrous tissue in close proximity to the prostate and the orifices of the ejaculatory ducts. This is referred to as imperforate rectum. It may remain in communication by a fine fistula which opens in immediate proximity to the normal orifices of the ejaculatory ducts and the *utriculus masculinus*. More rarely the fistula and Wolffian ducts open together at a higher level into the urinary system (Fig. I). These are familiarly known as recto-urethral and recto-vesical fistulae.

In the female, the rectal attachment or fistula to the urinary tract, at the level of Müller's tubercle, becomes incorporated in the actively proliferating and migrating epithelial vaginal bulbs. In changing

their location from the original level of Müller's tubercle below the bladder neck to the vestibule, these bulbs carry the rectum distally a varying distance. Sometimes the fistula is carried before the bulbs to the *fossa navicularis* in the vestibule, and in this paper it is referred to as a recto-vestibular fistula, but more frequently it is incorporated in the bulbs forming a fistula at any level into the posterior wall of the vagina (Fig. II). The orifice of the fistula into the posterior wall is usually wide in calibre, whereas that of the fistula into the *fossa navicularis* in the vestibule is small.

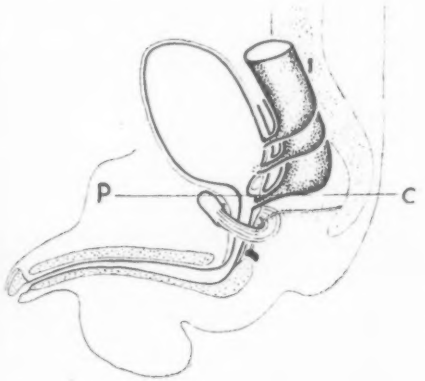

FIG. I. Showing the common level of the rectal fistula on the pubo-coccygeal line in the male and the more rare higher level fistula. The relation of the pubo-rectalis and levator diaphragm also shown. (newborn drawn to scale)

These vaginal deformities are easily recognized by the naked eye, though imperforate conditions of the rectum and recto-urethral fistula require the assistance of radiology.

SURGICAL ANATOMY AND PATHOLOGY OF THE DEFORMITIES

In order to elucidate these deformities and emphasize those features which have a direct bearing on the treatment of the condition, it is necessary to discuss:

(a) the anatomy of the *levator ani*,

(b) its abnormal development.

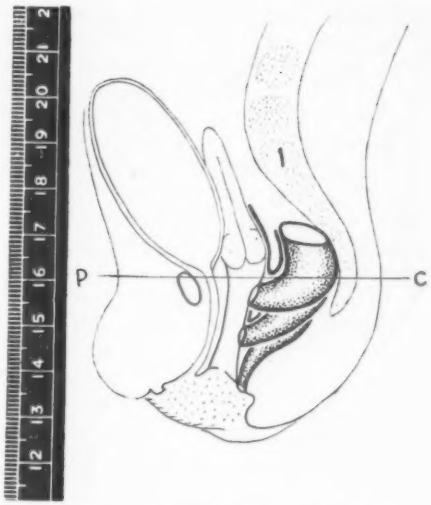

FIG. II. Showing the levels to which the rectal fistula is carried down the posterior wall of the vagina by the vaginal bulbs. The lowest is the recto-vestibular fistula.

It will be shown that, from both anatomical and radiological points of view, a line joining the pubis and sacro-coccygeal junction, the pubo-coccygeal line, is of special importance. The sphincters of the rectum also are of particular significance in the treatment of the deformities. All these features will be discussed in turn.

The normal levator ani

From the posterior surface of the body of the pubis and the white line as far back as the ischial spine, fibres of the *levator ani* sweep obliquely backwards medially and caudally in three sections. (Figs. IIIa and b).

One thin section sweeps obliquely across the membranous urethra and meets its fellow of the opposite side at the perineal body gaining delicate attachment to the urethra along its course.

A second strong sling gains attachment by its innermost fibres to the longitudinal muscle coat of the anal canal at the level of the anal valves. The remainder of its mass skirts the rectum and meets its fellow of the opposite side behind the rectum forming a strong collar of muscle, the *pubo-rectalis*. This band of muscle compresses the rectum, thus prolonging the narrow embryological

anal canal proximally to the upper border of the *pubo-rectalis* which, when contracted, forms the ano-rectal ring. The third section, the *pubo-coccygeus*, is continuous with the outer fibres of the *pubo-rectalis* and inserts in continuity with it into the ano-coccygeal *raphe* and the coccyx. These fibres blend with the *ileo-coccygeus* portion of *levator ani* which arises from the posterior part of the white line and the ischial spine.

with the anterior visceral sling which normally forms the *pubo-urethralis* or *vaginalis*. A strong collar of muscle is formed around the cloacal urethra or the vagina caudal to the terminal blind end or fistula of the hind-gut. In two specimens this muscle sling was confirmed microscopically by serial sections, to be striated, to be arranged as a sling and to be gaining intimate attachment to the urethral (cloacal)

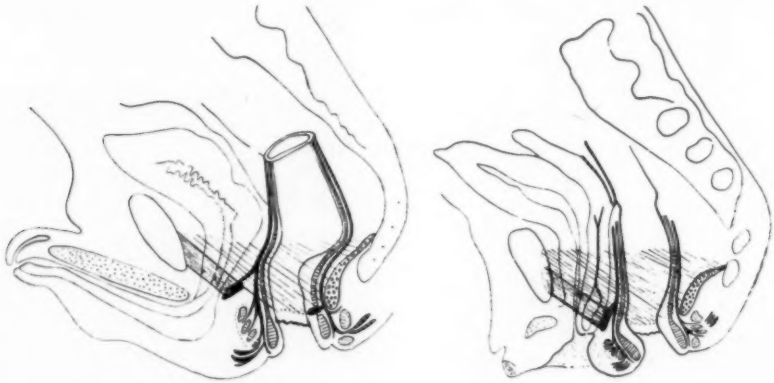

FIG. III a. FIG. III b.

Sagittal section of the pelvis (a) male, (b) female.

The main mass of the internal and external sphincters lies below the anal valves. The pubo-rectalis, lightly shaded area, gains attachment to the rectum along a line parallel with and a millimetre or two above the anal valves.

Heavy shading represents pubo-urethralis in (a) and pubo-vaginalis in (b).

By contraction of the two anterior slings, the urethra and rectum are compressed from side to side and the posterior wall of the rectum is elevated and angulated forwards. The contour of the *pubo-rectalis* assumes the shape of a collar, open in front, on a plane which is at right angles to the diaphragm. This diaphragm, on contraction, rises higher in the pelvis but plays no part in sphincteric control of the rectum.

In the female there is an additional sling called the *pubo-vaginalis*, which hugs the side walls of the vagina and inserts into the perineal body, and by its action the vagina is compressed from side to side.

Levator Ani in Imperforate Rectum With and Without Fistula

The fibres of the *levator ani* which form the *pubo-rectalis* in these deformities, fuse

canal in its lower part in the region of the perineal body. In one specimen of recto-vaginal fistula, microscopic section showed the *pubo-vaginalis* muscle lying below and posterior to the vagina in close relationship with the vaginal wall. In a case of recto-vestibular fistula with rudimentary vagina, serial sections confirmed the presence of a well-formed levator diaphragm and *pubo-rectalis* muscle. In the absence of the rectum, the diaphragm formed by the levatores muscle is elongated in the antero-posterior direction and the ano-coccygeal raphe is replaced by a raphe extending from the perineal body to the coccyx (Figs. I and IV).

Contraction of the sling and diaphragm causes the collar-like compression of the cloacal urethra or vagina and raises the flat surface of the diaphragm to the level of

origin of this muscle in the pelvis—a level a little below the pubo-coccygeal line (Fig. Xa).

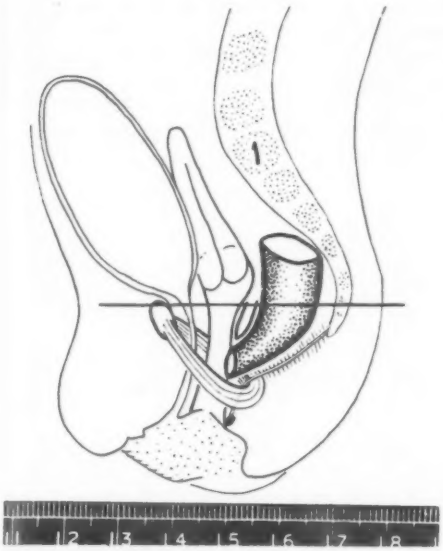

FIG. IV. Recto-vaginal fistula showing relation of pubo-rectalis sling and *levator ani* diaphragm to the fistula (common to all three in Fig. II).

Spinal Abnormalities and Influence on the Formation of Levator Ani

In the 29 cases of imperforate rectum with and without fistulae, the sacrum and coccyx were normal in 9, abnormal in 17 and unknown in 3. The abnormalities consisted of hemi-vertebrae (2 cases), recurved sacro-coccygeal segments (2 cases), sacral *spina bifida* (1 case), absent coccyx (3 cases), and deficiencies in the terminal end of the spine (S.5, 1 case; S.4, 6 cases; S.2, 2 cases). Of the seventeen abnormal spines, post-mortem examinations were made in twelve.

Correlating the clinical and surgical findings with the pathological anatomy of the specimen, it appears that the *levator ani* musculature may be strongly developed in the absence of the fifth sacral segment, thinly developed in the absence of S.4, and totally deficient in absence of S.2 (one case).

It must be understood that the assessment of deficiencies of the spine can be only approximate unless all vertebrae are visualized by X-ray examination. Total numbers of vertebrae are not known in this series. For the purpose of clinical management of these cases, an approximate estimate is helpful, though full spinal vertebrae counts are probably an advantage.

It is usually possible to draw in the pubo-coccygeal line in cases of lesser deficiencies of the spine by making the necessary allowance for the absent vertebrae. However, in the gross cases, it is found that the hind-gut frequently lies entirely above the false pelvis, and the line is of no clinical value.

TABLE 1

Age and Sex of 25 Pelves

Age	Male	Female
4th to 9th Foetal Month	10	11
Infants and Children	3	1

PUBO-COCCYGEAL LINE

The normal anatomy of the organs was studied in 25 pelves (Table 1).

The anatomical relation of the bladder neck, the external *os* of the *cervix uteri*, the *verumontanum*, the peritoneal reflexion from the rectum to the prostate, and the third fold of Houston were found to be close to a line drawn on the bisected specimen from the upper surface of the symphysis pubis to the sacro-coccygeal junction (Fig. V). The level of the pouch of Douglas in the female varies considerably in the level of the peritoneal reflexion. In nine cases it was on the same level as this line, but in one case it was considerably higher and in two cases it was much lower (Fig. VI).

This line can be traced on lateral X-rays of the pelvis in newborn babies and in older children. The X-ray tube should be centred over the greater trochanter of the femur. The landmarks are, (i) the upper border of the *symphysis pubis*. The region of the

symphysis is accentuated and recognizable by the overlapping of the pubic ramus and body which, when superimposed by the ossific centre from the opposite side, casts a relatively dense shadow. The ossification of the pubis begins at the second foetal month under the acetabulum and extends along the superior ramus to the body and at birth it is as shown in Fig. VII.

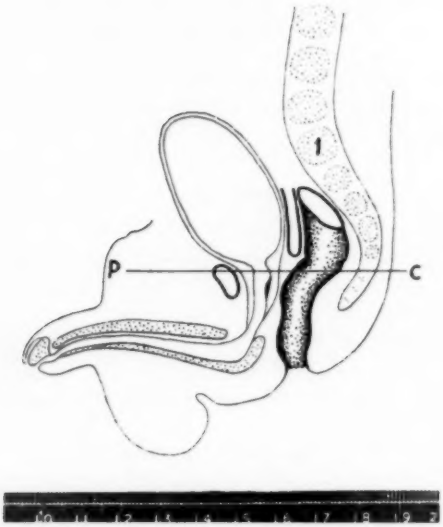

FIG. V. Demonstrating the pubo-coccygeal line and the structures related to it in the male—upper border of symphysis, peritoneal reflection and Houston's fold. The *verumontanum* is immediately below it.

(ii) The last piece of the normal sacrum. The centre for this fifth piece appears at approximately the fourth month of foetal life and is showing as a small ossific ring at birth. The sacro-coccygeal junction which is the posterior landmark, is immediately distal to it. The coccyx begins to ossify at approximately the second to the fifth year and is therefore not visible till some months after birth.

(iii) This is a thin lead marker placed on the lower lip of the crease line which is constantly present in the newborn, marking the upper surface of the body of the pubis. It is the lower of two creases and its position is confirmed by palpation of the pubis with the finger.

All three landmarks on the lateral film of the pelvis are joined to form the pubo-coccygeal line. This is a fixed line about which the pelvic structures can be assembled to "anatomize" the X-ray appearance. This line provides a satisfactory clue to the anatomical level of abnormalities of the rectum and anus.

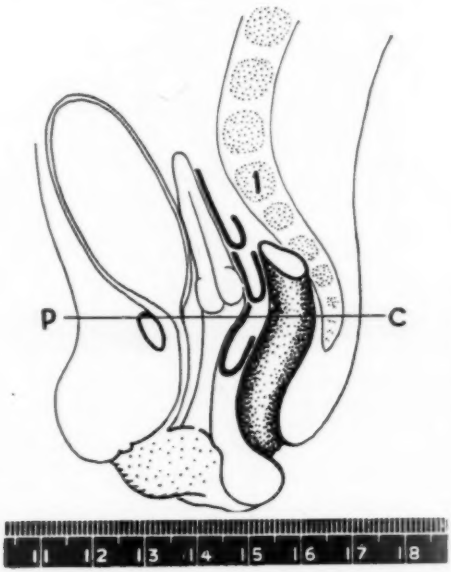

FIG. VI. Showing the levels to which the pouch of Douglas descends in the normal pelvis.

RADIOLOGICAL IDENTIFICATION OF TYPES OF RECTAL DEFORMITIES

In order to demonstrate the terminal blind end of the gut in cases of congenital absence of the middle rectal segment in the newborn, the Wangensteen-Rice (1930) principle is employed. True lateral views of the pelvis are taken with the child inverted. The gas in the bowel rises to the top displacing the meconium from the terminal end. X-ray examination demonstrates the smooth outline of the gut distended by the gas which stands out in contrast to the solid tissues of the pelvis.

FIG. VII. To demonstrate ossific centres and bony landmarks in newborn full-time baby. The upper end of the pubis is denser than the remainder due to superimposed bone. The coccyx is not ossified.

Certain precautions in interpretation are necessary:—

(i) Until some degree of distension of the abdomen has developed, and this may not occur before 24-36 hours, the gases may be insufficient in amount at the terminal end of the gut to displace all the meconium, rendering the interpretation unreliable.

(ii) The outline of the terminal gut as shown by the gas shadow must be smooth, rounded or beaked, and well distended.

(iii) In the presence of a wide or short fistula, for example, to the vagina or skin, gas in the terminal end escapes and the accumulated meconium plug renders this contrast method impractical. In most of these cases the level of the fistula can be visualized by the naked eye and the type of deformity identified with a speculum.

The lumen of the recto-urethral fistula, however, is so small and long that it is easily clogged with meconium which prevents the escape of gases for long periods in most cases, and the gas outlines of the terminal rectum are reliable.

The contrast X-ray technique using lipiodol is used to determine the level of the terminal end of the gut in babies who have gained relief by colostomy. The lipiodol is introduced through a catheter in the distal limb of the colostomy and the films of the pelvis are taken with the infant erect.

Relation of Abnormalities to Pubo-coccygeal Line

In male rectal deformities, the hind-gut shadow most commonly lies closely related to the pubo-coccygeal line. This is so for both the imperforate rectum and the recto-urethral fistula (Fig. VIII). More rarely the terminal gut, gaining attachment to the posterior wall of the bladder, can be identified at a higher level. A recto-urethral fistula in the male below this line but above the bulb was not seen in this series.

In the female, recto-vaginal fistulae open into the posterior wall of the vagina from the level of the pubo-coccygeal line at any level as far distally as the outlet and *fossa navicularis* in the vestibule. Rectal fistulae above the level of this line open in the midline into the bladder neck or trigone region. In these cases the right and left Müllerian ducts remain separate, penetrating the musculature of the bladder neck or bladder independently to either side of the fistula. The female counterpart of the male recto-urethral fistula on the pubo-coccygeal line is a similar condition altered only by the enlargement of the *utriculus* into a short double vagina and bicornuate uterus. In the four cases studied, the deficiencies in the spine rendered accurate definition of the pubo-coccygeal line impossible and the level was approximate only.

EMBRYOLOGICAL SUBDIVISION OF THE CLOACA

From the above descriptions it appears that this line represents an important embryological level. Wood Jones (1904) emphasized the frequency with which recto-urethral fistulae open at the level of the *verumontanum*. His theory of the post-allantoic outgrowth from the hind-gut to form the intermediate segment of rectum

explains many of these rectal anomalies, particularly the common recto-urethral fistula but in many subsequent studies in human embryology this theory has not been confirmed.

below the level of the Müller's tubercle. In so doing the receding Wolffian duct orifices are carried in the mesenchyme from their original lateral to para-central positions. The mesenchyme indents the sides of the

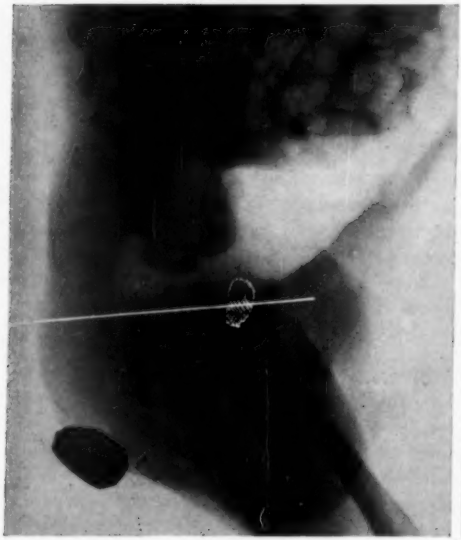

FIG. VIIIa. Recto-urethral fistula. Lateral X-ray of pelvis showing gas in terminal gut extending to the pubo-coccygeal line. The marker is on the skin dimple.

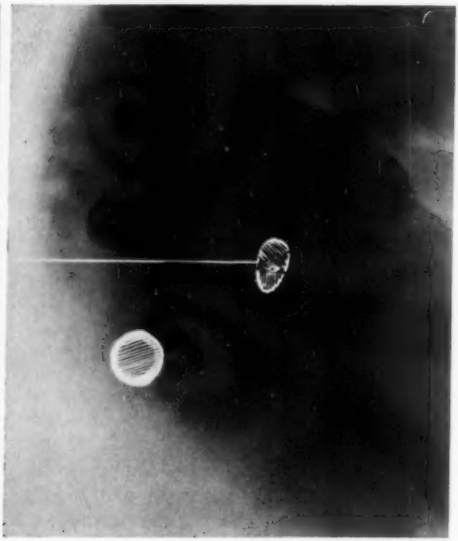

FIG. VIIIb. Imperforate rectum, showing lipiodol extending to pubo-coccygeal line. The rounded marker lies in a well-developed anal pit.

The theory of the uro-rectal septal downgrowth as a means of subdividing the cloaca is strongly favoured by embryologists (Hamilton, Boyd and Mossman, 1945). If this were the mechanism, the commonest form of fistula in the male would be a fistula at the level of the triangular ligament. This is the site of the so-called cloacal duct which is the last part to close. The extreme rarity of such a fistula at this level and in the membranous urethra, and the frequency of the fistula at the level of the pubo-coccygeal line, suggest that the subdivision of the cloaca below this line takes place by a different mechanism from that which cleaves the gut from the urinary tract above this point.

It is suggested here that the inward surge of mesenchyme from the sides of the embryo towards the midline completes the division cloacal cavity as described by Keibel and Mall (1912) and illustrated by Bradley Patten (1947), dividing the compartment below the pubo-coccygeal line. The uro-rectal septum above this level is formed as the plane of cleavage between the *allantois* and the hind-gut as a result of the elongation and rotation process of the tail and hind end of the embryo.

The failure of subdivision by the lateral ingrowths accounts for the common abnormality of recto-urinary fistula in the male at the level of the pubo-coccygeal line. The urethra, actually below the fistula, is the undivided cloaca. In the female, this fistula, being incorporated in the actively proliferating and migrating epithelial vaginal bulbs, is carried in the posterior wall of the vagina to a lower level.

SPHINCTERS IN RECTAL ANOMALIES

In the investigation of these abnormalities, the sphincters were studied in order that they could be used to best advantage in the reconstructive surgical procedure. The *levator ani* musculature has already been described. The internal and external sphincters were rudimentary.

(i) Internal sphincter muscle. Serial sections in one case confirm the macroscopic impression obtained from three cases that the internal sphincters are lacking in male cases of recto-urethral fistula. Microscopic examination of one case of recto-vaginal fistula indicates that there is slight thickening of the terminal ends of the muscle coats of the rectum, but this was considered to be due to obliquity of section of the muscle. This thickening was not noticeable on macroscopic examination and bore no resemblance to the normal internal sphincter.

(ii) External sphincter muscle. One case of recto-urethral fistula, which was completely dissected, demonstrated clearly a sagittal sail-like velum of voluntary muscle arising from the perineo-coccygeal *raphe* and inserted directly into the anal pit (Fig. Xb). Its action was to cause dimpling of the anus, a sign which is sometimes fallaciously believed to indicate a well-formed external sphincter.

Serial sections of one case of recto-urethral fistula confirmed the presence of this sail-like *velum* of voluntary muscle lying in the sagittal plane, though no actual insertion into the skin was apparent as in this case there was no anal dimple.

The internal and external sphincters in the rectal deformities examined were of negligible value. The *levator ani* musculature is well-developed and provides the main sphincter mechanism available to surgeons in reconstructive rectal operations.

OPERATIVE TECHNIQUE IN RECTO-URETHRAL AND RECTO-VAGINAL FISTULA

A transverse defunctioning colostomy is performed to deflate the bowel and further operation on the rectal deformity is delayed for a year to allow the baby to increase in size.

The approach to the blind end is possible—

(a) from the perineum,

(b) combined perineal and abdominal approach, or

(c) through the sacro-coccygeal route.

The sacro-coccygeal route is chosen because the perineal operation is likely to cause damage to the *levator ani* muscle. The combined perineo-abdominal method may direct the new anal canal posterior to the sling-like collar which lies anteriorly in close relationship to the pubis. The rectum may then perforate the levator diaphragm in its non-sphincteric portion.

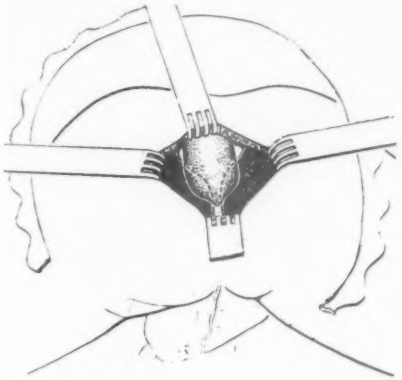

FIG. IX. Showing exposure of the supra-levator space through a natal cleft incision with disarticulation of the sacro-coccygeal junction.

It has been found that through the sacro-coccygeal approach, the rectum can be mobilized sufficiently to bring it to the perineum when the blind end reaches to the pubo-coccygeal line or lower. Otherwise preliminary mobilization is necessary by abdominal incision.

A midline incision is made in the natal cleft, centring over the sacro-coccygeal junction. A transverse incision is then made through the cartilage of this articulation and the incision is widened laterally for about a centimetre on each side.

A retractor is then placed in the opening separating the coccyx from the terminal sacral vertebra (Fig. IX).

This brings the posterior aspect of the pelvic rectum, clothed in its fascia, into

view in the supra-levator space. This fascia is incised and stripped with the finger from the rectum.

In the male

The attachment of a fistula to the urinary tract, emerging from the antero-inferior aspect of the gut (Fig. Xa) is then cleared with angled forceps. The rounded end of the rectum is incised and the fistula is inspected from within. The fistula is clamped, ligated and divided. The detached rectum is then cleared and mobilized proximally by finger dissection.

FIG. Xa. Drawing to demonstrate the relation of the pelvic diaphragm to the "recto-urethral" fistula. The *symphysis pubis* has been divided and opened out to demonstrate the pubo-rectalis sling.

A sound is then passed into the urethra to demarcate its course. The beaks of a right-angled forceps are introduced through the supra-levator space and passed forwards as far as the urethra (Fig. Xb). At this point the beaks are directed caudally parallel with the sound for a short distance and then posteriorly to the skin of the perineum at the normal site of an anus. The object of this manoeuvre is to ensure that the forceps keep well within the levator sling. A wide ellipse of perineal skin is then removed over the eminence made by the beaks of the forceps. A piece of tape is engaged in the jaws of the forceps in the perineum and is withdrawn through the pelvis, inside the sling of the levator and out through the posterior incision. By gentle traction on both ends the levator sling is elongated backwards.

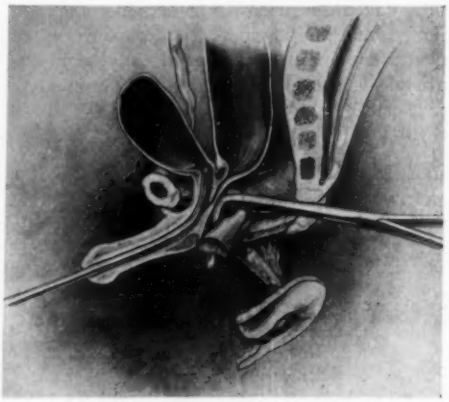

FIG. Xb. Demonstrating the principle of reconstruction of the rectum conserving the "pubo-rectalis" muscle as the new sphincter mechanism. Posterior traction on the sling makes room for the new anal canal — Fig. Xc. This diagram also shows the thin sail-like *velum* of voluntary muscle, the rudimentary external sphincter, inserting into the anal dimple.

The terminal end of the mobilized rectum is now directed to the perineal opening through the enlarged muscular sling (Fig. Xc).

The rectum is sutured to the skin, the tape is then withdrawn and the posterior incision is closed in layers.

In the female

(a) Recto-vestibular fistula.

As stated before, this anomaly must be distinguished from the covered anus with projection of the anal lumen anteriorly to the verge of the vestibule. This is done by observing the direction a probe takes when introduced into the orifice of the fistula. In the recto-vestibular fistula, the probe passes deeply in a cranial direction, whereas in the covered anus the probe can be passed directly backwards under the skin of the perineum.

The recto-vestibular fistula is best treated by a superficial midline posterior incision of the skin, directly posterior to the orifice of the fistula, sufficient to permit the entry of the sounds approximating size 18Fr. This is dilated during the next three weeks

to the calibre of the fifth finger. Subsequently, the parent dilates the passage daily until the anus is supple and of adequate calibre. Two patients treated in this way developed efficient sphincter control.

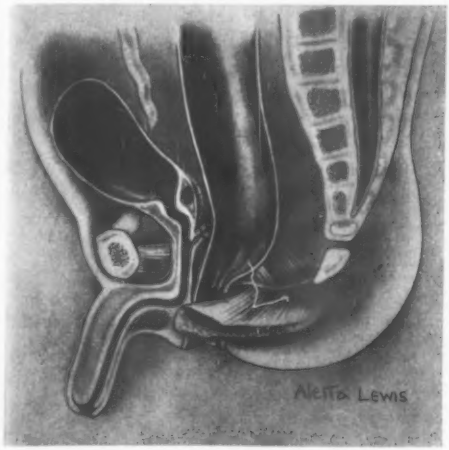

FIG. Xc. Drawing showing the completed operation with the pubo-rectalis encircling the new anal canal.

(b) Recto-vaginal fistula.

A similar technique to that of the recto-urethral fistula in the male is used for recto-vaginal fistula.

The supra-levator space is exposed through the same incision, the recto-vaginal fistula is dissected free and divided on the wall of the vagina. The deficiency in the vagina is then oversewn with catgut. A sound is passed into the lumen of the vagina and the same manoeuvre as for the male is carried out with the angled forceps. A tape is withdrawn with the forceps through the supra-levator space and the pubo-vaginalis muscle is eased backwards to provide a space within the sling through which the rectum is directed to the perineum.

Operations using this technique were performed on four children.*

*The author is grateful to Mr. T. Twistington Higgins and Mr. Denis Browne at The Hospital for Sick Children, London, to Mr. Naunton Morgan at St. Mark's Hospital, London, and to Mr. Whitaker at the Melbourne Children's Hospital, for permission to assist them in the operations on these cases, and to report them in this paper.

Case 1

G.C., aged 1 year (Mr. Denis Browne's case). Recto-urethral fistula, opening into the *verumontanum* situated at the normal level in the pelvis. The fourth and fifth sacral vertebrae and the coccyx were deficient. A colostomy was performed soon after birth. At approximately 1 year, the new anal canal was constructed according to the principle described in this paper. No complications occurred, but the follow-up disclosed persisting incontinence, which may be accounted for by a weakness of the *levator ani* musculature associated with the spinal deformity.

Case 2

A.H., aged 5 years (Mr. Higgins' case). Imperforate rectum which ended blindly in the vicinity of the base of the prostate. Both ureters were considerably dilated. Soon after birth, a colostomy, and later a laparatomy, was performed. At five years of age, the new anal canal was constructed. The mucocutaneous function of the new anus formed a stricture and remained narrow in calibre. This child gained some measure of continence and rectal sensation, though a slight leakage of the more fluid motion occurred several times a day.

Case 3

S.W., aged 4 years (Mr. Naunton Morgan's case). Recto-vaginal fistula, which entered the posterior wall of the vagina a little below the level of the external os. A colostomy had been made in infancy. At 4 years, the fistula was repaired and the new anal canal was fashioned. This child developed continence (personal communication from Mr. Naunton Morgan), though she could not distinguish between faeces and flatus.

Case 4

R.W., aged 2 years, 10 months (Mr. Whitaker's case). Imperforate rectum with blind end lying at level of the prostate. The fourth and fifth sacral vertebrae and coccyx were absent. On 2nd day of life, a colostomy was performed, and later the abdomen was explored with a view to constructing a new anal canal without success. At 19 months, a new anal canal was constructed by the above technique. Subsequently it was necessary to correct a skin stricture of the new anus by rotation of a skin flap into the orifice. This child, after one year, is incontinent, though more easily managed than with the colostomy. Occasionally a little urine leaks through the new anus. The incontinence may be accounted for by the spinal deficiency and an associated abnormality in the *levator ani musculature.*

Though too few cases have been treated by this method, and though the results are not enough to prove as yet the success of the procedure, it is considered that the design of this operation uses to the best advantage the muscles, which are available in this class of deformity, in the construction of a sphincter mechanism.

SUMMARY AND CONCLUSIONS

1. Anal and rectal deformities are usually clearly distinguishable by clinical and radiological methods. This study is confined to the rectal group.
2. Imperforate rectum with or without a urinary or vaginal fistula is a standard pattern deformity resulting from failure of subdivision of the endodermal cloaca.
3. The pubo-coccygeal line joining the upper surface of the pubis with the sacro-coccygeal junction marks the level of the bladder neck, the external *os* of the *cervix uteri*, Houston's third fold of the rectum, and in the male, the lowest limit of the peritoneal reflection. In the female the pouch of Douglas most commonly extends to this level, but may be higher or lower in the pelvis.
4. The origin of the *levator ani* musculature is on a slightly lower level than the pubo-coccygeal line.
5. The terminal end of the hind-gut in imperforate rectum deformities extends to or proximal to this line. In the female, the hind-gut may become incorporated in the actively proliferating vaginal bulbs which migrate from the level of this line to the perineum, carrying the rectum as a fistula in its posterior wall to a variable position in the pelvis below the line.
6. The abnormal *levator ani* musculature is formed as a diaphragm across the pelvis a short distance below the pubo-coccygeal line and is arranged as a sling around the viscera which traverse the pelvis to the perineum. This sphincteric sling of muscle lies below and anterior to the free terminal end of the hind-gut and the fistula when present.
7. Major deficiencies in the sacrum, involving the higher sacral vertebrae, are accompanied by corresponding lack in development of the *levator ani* musculature.
8. The internal and external sphincters in these imperforate rectum deformities are very rudimentary and of little value in reconstructive surgical procedures.
9. The level of the distended terminal end of the hind-gut can be demonstrated by appropriate X-rays, in all but the wide calibre fistulae which permit the free escape of gases from the bowel. The structures in the pelvis can then be "anatomized" in relation to the pubo-coccygeal line.
10. The sacro-coccygeal approach to the hind-gut and fistula in the supra-levator space is used in the reconstructive surgery of imperforate rectum.
11. Provided the rectum descends to the level of the pubo-coccygeal line, sufficient mobilization is possible through this approach to enable the terminal end to be directed within the sphincteric sling of the *levator ani* musculature to the perineum. This sling now forms the *pubo-rectalis* which is the main sphincter of the newly constructed anal canal.

ACKNOWLEDGEMENTS

This study was begun at The Hospital for Sick Children, Great Ormond Street, London, under the invaluable guidance of Mr. Denis Browne, from whose clinic a large part of the material was obtained and Dr. Martin Bodian, for whose help I wish to acknowledge my sincere thanks. I am grateful to Mr. T. T. Higgins and other members of the staff for permitting me to co-operate with them on their cases. The work has been continued at the Children's Hospital, Melbourne, and my thanks are due to Mr. Whitaker for permission to use some of his material, and to Dr. J. Perry for assistance in the Pathology Department. Professor S. Sunderland has given valuable advice and much technical assistance in the preparation of serial sections of the foetal and newborn pelvis. Throughout this period of study from 1947 to 1952, generous grants have been made available by the Nuffield Foundation.

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POLYCYSTIC DISEASE OF THE LIVER

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WHERE presented as a clinical problem, polycystic disease of any viscus is sufficiently rare to arouse interest. Except when this occurs in the kidneys and produces eventual renal failure, it is seldom that cystic disease gives rise to symptoms so that discovery of the condition is usually made post-mortem (Burns, 1925). Particularly is this true of non-parasitic cysts in the liver where, invariably, there remains an amount of normal liver tissue adequate for physiological requirements. Moreover, when cystic disease affects the liver, the same condition is often found in the kidneys, the pancreas or the lungs.

The following case is presented because, not only did its symptoms pose a diagnostic problem, but also the cystic disease was confined to the liver.

FIG. I. Skiagram showing large cyst of right lobe of liver. The two larger arrows indicate the small calcified plaques in the cyst wall.

Case Report

The patient was a healthy looking married woman aged 55 years. During some years prior to 1951 she had experienced attacks of dull pain in her upper middle and right abdomen. Each attack was never more than a few hours in duration and there was never any nausea or vomiting. The attacks occurred intermittently at irregular intervals of several weeks. Apart from these attacks of pain she also complained of flatulent dyspepsia with an intolerance for highly seasoned foods. Her doctor had regarded these features as being indicative of mild chronic cholecystitis and referred her for surgery when her symptoms became aggravated over a period of six weeks. When she was examined at this time, the salient feature was a large, round swelling palpable within the right side of her abdomen. This swelling merged with the liver above and it was observed to move synchronously with diaphragmatic respiration. It gave the sensation of being elastic and tense, but its position well towards the right flank, and its size about that of a tennis ball, seemed inconsistent with a diagnosis of enlarged gall-bladder. Moreover, it was not tender. The possibility of its being a hydatid cyst of the liver was postulated.

FIG. II. Photograph of large cyst from right lobe of liver showing thin translucent but vascular wall.

The hydatid complement fixation and the Casoni tests were both negative. An X-ray cholecystogram showed the outline of a normal sized gall-bladder which functioned well. Lying lateral to the gall-bladder and extending downwards from the right lobe of the liver was seen a large cyst, in the wall of which, at its lower pole and its medial boundary, were small areas of calcification (Fig. I).

Operation

At operation on the 12th September, 1951, (performed by N.W.) access was gained by a transverse abdominal incision above the umbilicus extending from the midline to the right flank below the costal margin. On opening the peritoneum there presented a thin walled translucent cyst of the right lobe of the liver. It was roughly spherical, about 8 cm. in diameter, and extended downwards almost as far as the crest of the ilium. It was quite unlike the hydatid cyst which had been expected. Adjacent to it were two small, similar, serous cysts. Further search revealed many more cysts distributed in various parts of the otherwise normal liver substance. High on the anterior surface of the right lobe were three small cysts, 2 cm. or more in diameter, almost buried within the liver. In the quadrate lobe was another, 4 cm. in diameter, and in the same lobe one more cyst of slightly smaller dimension abutted upon the *porta hepatis*. In a somewhat diminutive left lobe of the liver were numerous cysts of varying size, dwarfed by one large one 6 cm. in diameter, which had expanded between the layers of the falciform ligament and to which part of the great omentum was firmly adherent. The gall-bladder and all the other abdominal viscera were normal in all respects.

FIG. III. Photograph of cysts from left lobe of liver with adherent omentum. The cut surface shows the largest cyst to be filled with haemorrhagic debris.

Commencing with the largest cyst in the right lobe of the liver so as to give better access to the remainder, all the cysts were totally excised by cutting through the liver tissue surrounding them. This was necessary because of the entire absence of a capsule to any of them. The left lobe of the liver required total extirpation together with the greater portion of the falciform ligament and some omentum. Bleeding was controlled by ligation of individual large vessels and by mattress sutures through Glisson's capsule. A split rubber tube drain to the sub-phrenic region was inserted as the abdomen was closed and three days later, by which time oozing had ceased, this drain was removed. Post-operative transient collapse of the lower lobe of the right lung responded satisfactorily to appropriate treatment and was the only complication. The patient made a good recovery and left hospital on the twentieth day after operation. She has remained free from all her previous symptoms up to the present and is in excellent health.

FIG. IV. Photomicrograph of a cyst wall showing epithelial lining, fibrosis, an inflammatory cell infiltration and several small bile ducts. (Van Gieson, x 200)

Gross Specimen

Examination after fixation showed six separate portions of liver tissue containing cysts. The large cyst from the right lobe measured 7 cm. in diameter (Fig. II). The large cyst in the left lobe measured 5 cm. in diameter and had omentum adherent to it. This cyst contained chocolate-coloured haemorrhagic debris and its fibrous wall was up to 0.1 cm. in thickness (Fig. III). All the other cysts had thin, translucent walls and contained clear, colourless fluid.

Histology

Microscopically the cysts have thin, collagenous, fibrous, tissue walls and some are lined by flattened epithelial cells (Fig. IV). In other cysts the lining comprises a zone of young fibroblastic tissue the inner cellular layer of which resembles a mesothelium (Fig. V). It would appear that, in these, the original epithelial lining has been destroyed by pressure and replaced by fibrous tissue. The portal tracts surrounding the cysts show some fibrosis and an infiltration by chronic inflammatory cells. Apart from a moderate degree of patchy, mid-zone, fatty infiltration, the liver parenchyma is normal. There is no evidence of echinococcus infestation and the cysts must be regarded as the result of some developmental defect.

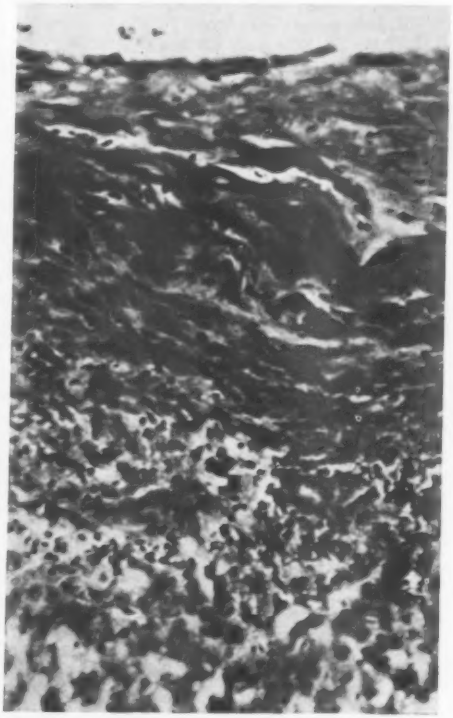

FIG. V. Photomicrograph of another cyst wall showing lining of fibroblastic tissue the inner layer of which resembles mesothelium. This young fibrous tissue rests on an older, folded, hyalinised collagenous layer in the outer zone of which are inflammatory cells. (Van Gieson, x 200)

DISCUSSION

Solitary retention cysts of the liver may be acquired as a result of bile duct obstruction but multiple cystic disease is developmental in origin although the mechanisms concerned are disputed (Rolleston and McNee, 1929; Norris and Tyson, 1947). In 1913 Boyd noted that, in the reported cases of multiple cysts of the liver, females predominated over males by three to one. The incidence of the condition has been given by Ackman and Rhea (1931) as 11 cases in 6,141 autopsies (0.18 per cent.). These authors observe that symptoms, when they occur, simulate those of chronic cholecystitis. These symptoms are due to complications such as sudden undue pressure on the extra-hepatic bile ducts, acute intra-cystic haemorrhage, or secondary infection. In our case it seems likely that repeated haemorrhage into the large cyst of the left lobe was the reason for her attacks of vague dull pain in the upper abdomen. Displacement of and pressure upon the adjacent colon by the large cyst of the right lobe may explain the symptoms of flatulent dyspepsia.

In countries such as Australia and New Zealand where hydatid cysts of the liver are not uncommon, it is proper that such will be the pre-operative diagnosis in cases where clinical and radiological findings indicate cysts to be present.

However rare cases of non-parasitic cysts may be, they are of importance, because surgeons accustomed to operate for hydatid cysts may find themselves unexpectedly confronted with this entirely different problem, treatment of which may prove hazardous.

Up to 1923 Jones found reports in the literature of 61 cases treated surgically and by 1937 the number had risen to 226 (Davis). In two cases, that of Von Haberer in 1909 and that reported in 1948 by Clagett and Hawkins, the entire left lobe of the liver was excised, as it was found necessary to do in our case. In some of the cases the surgery has amounted to no more than the taking of a biopsy specimen; in others the cysts have been marsupialized and packed with gauze which is later withdrawn. The latter procedure may be the only feasible method when anatomical difficulties preclude total excision which is the method of choice whenever possible. It is generally agreed

that puncture and evacuation of the cysts is quite useless because they persist and continue to grow in size.

SUMMARY

A case is described in which clinical symptoms were caused by multiple non-parasitic cysts of the liver. The other abdominal viscera were normal. The surgical treatment is discussed with reference to the experience of other surgeons.

ACKNOWLEDGEMENTS

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HAMILTON RUSSELL MEMORIAL LECTURE

PULMONARY OEDEMA

CLINICAL IMPLICATIONS OF LABORATORY EXPERIMENTS

By F. C. COURTICE

Kanematsu Memorial Institute of Pathology, Sydney Hospital, Sydney

THIS evening we have assembled to do honour to the memory of one of our great surgeons and a founder of this College. Hamilton Russell came to Melbourne from England in 1889 after training as dresser and house surgeon to Lister. His work in Melbourne from that time until his death in 1933 is well known to you all, and has been acclaimed throughout the world.

At first I wondered whether it was appropriate for an experimenter and physiologist to give a lecture in memory of this surgeon before an audience of the leading surgeons in Australia. When I realized, however, that Hamilton Russell lived and studied medicine as a young man during that era of scientific progress when the laboratory experiments of scientists were paving the way for the revolution in the art and practice of surgery, I felt much happier in accepting the honour that your College has bestowed upon me. No doubt his early training with such a man as Lister and at such a time in the history of surgery, bred in Hamilton Russell that scientific spirit of practical enquiry and originality which he possessed.

I have chosen the subject of this lecture because it is an old one which has fascinated and puzzled generations of medical men, and also because I, myself, have been interested in and have worked on various aspects of the physiology of the lungs for several years. The subject I thought also appropriate because there is now required of surgeons an increased understanding of the physiology of the body fluids.

Some surgeons with whom I have discussed this problem tell me that pulmonary oedema is rare compared with other pulmonary complications and that the risk of giving even large transfusions is little. But, on occasions, acute pulmonary oedema does intervene post-operatively and may even be the primary cause of death. I remember a few years ago a surgeon coming from the post-mortem room with a grossly oedematous lung in his hand and expressing surprise and astonishment that pulmonary oedema should have suddenly caused the death of his patient who was doing so well post-operatively. No single cause of the sudden oedema could be postulated at the time although the patient had received considerable transfusions.

TABLE 1

The degree of pulmonary oedema in 72 post-operative deaths at Sydney Hospital in 1949-1952

Grade of oedema	Number of cases	Percentage
0	28	39
1	24	33
2	10	14
3	10	14
	72	100

It is often very difficult to assess accurately the extent of pulmonary oedema both clinically before death and also after death in the post-mortem room, especially in patients suffering from other chronic pulmonary conditions. Nevertheless, in collaboration with one of my colleagues, Dr. Bruce Hurt, I have investigated the pathology records over the last three years of all surgical cases dying in Sydney Hospital within a fortnight of operation, the majority (84 per cent.) within the first week. From the macroscopic and microscopic appearances of the lung, the degree of oedema has been graded 0, 1, 2

and 3. Table 1 shows the distribution in the group of 72 such cases. This table suggests that pulmonary oedema does occur in some of these cases, and, even if not the primary cause of death, it might be a contributory factor.

days before death and the degree of oedema at death. This relation is also shown in a similar group of cases in 1934-36 when postoperative transfusions were less common than to-day. It is evident that there is no great difference between the incidence of pul-

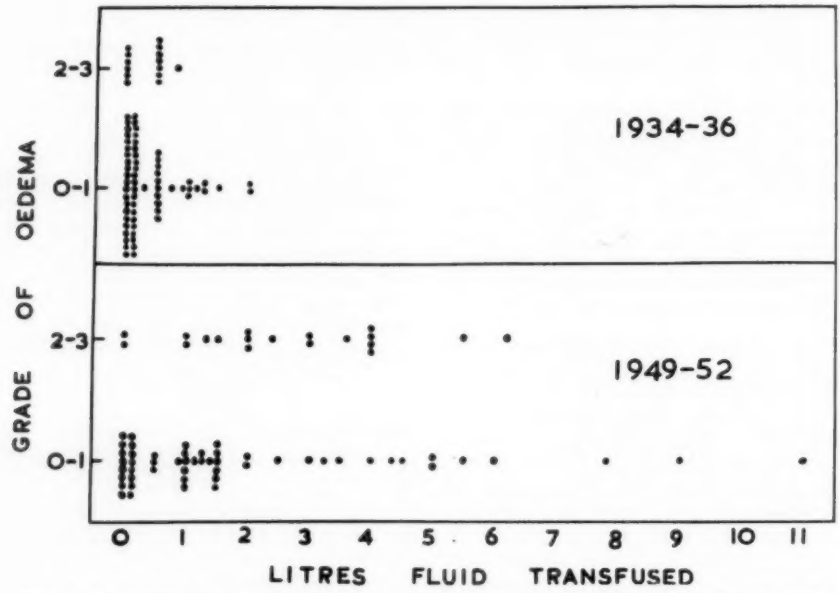

FIG. I. The relation between the degree of pulmonary oedema at death and the volume of fluid transfused during the last 3 days before death in two groups of surgical cases at Sydney Hospital, during the periods 1934-36 and 1949-52.

The problem of sorting out the cause or causes of pulmonary oedema in these cases is more difficult. An examination of the heart pathology showed that there was hypertrophy, fibrosis or pericarditis in 11 out of 28 in grade 0, 12 out of 24 in grade 1, 9 out of 10 in grade 2, and 6 out of 10 in grade 3. The lung pathology was variable, but no clear distinction could be drawn between the different groups. Both of these factors might influence pulmonary oedema (Cameron, 1948), but, although there is a possible relation between the heart pathology and the extent of the oedema, it is difficult to assess in this group of cases the extent of oedema due to these causes. Lastly, the amount of fluid transfused might have influenced the degree of oedema. In Fig. I is depicted the relation between the amount of fluid given intravenously in the last three

monary oedema in the 1934-36 group when very little fluid was transfused and that in the 1949-52 group in which considerable amounts of fluid were sometimes given intravenously, mainly glucose-saline, but also blood, serum and normal saline.

This is but a very brief survey of a group of cases taken from our records at Sydney Hospital. What it shows is that in a group of surgical patients, even when we know the full clinical course of the case, and the macroscopic and microscopic appearances of the lungs as well as of all the other organs of the body, it is difficult to get very far in our basic thoughts of why some patients develop pulmonary oedema. It is equally puzzling, on studying some records, why the lungs of others do not become grossly oedematous.

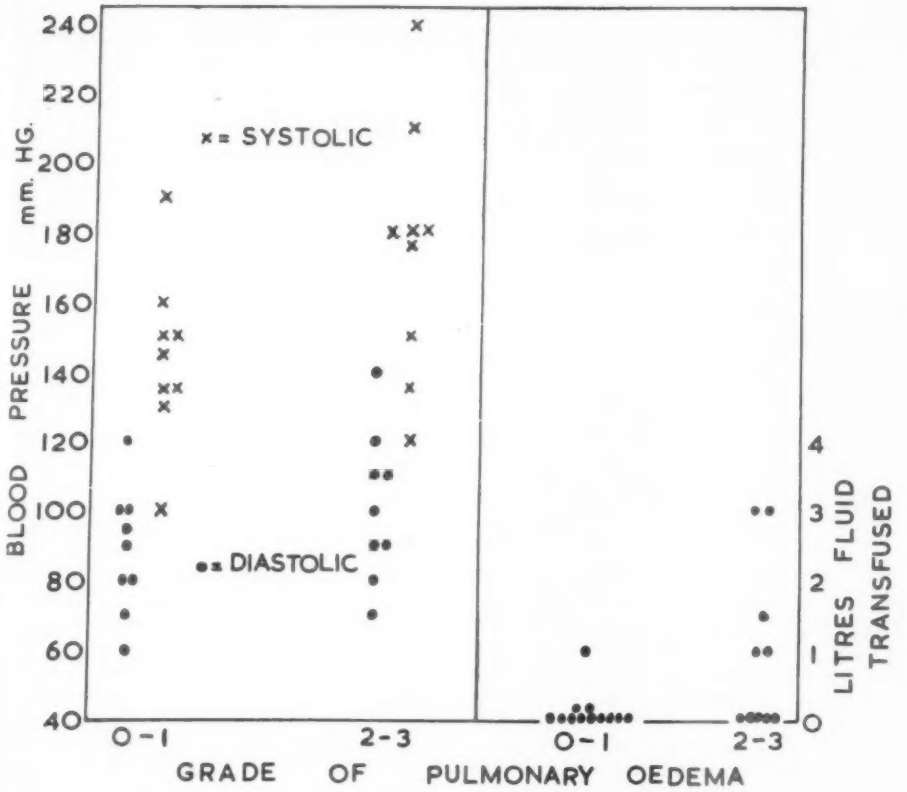

FIG. II. The relation between the degree of pulmonary oedema at death and the blood pressure and volume of fluid transfused in a group of cases from Crown Street Women's Hospital, Sydney.

In another group of clinical cases from Crown Street Women's Hospital, I have attempted to analyze the incidence of pulmonary oedema, in conjunction with Dr. S. Meares and the pathologist of the Hospital, Dr. Murray Moyes. This is a group of women, all of the same age group, who have died during pregnancy or just after parturition. In fairness to the staff of this hospital, I must emphasize that the majority of these cases entered hospital *in extremis*. Dr. Moyes has assessed the degree of pulmonary oedema from the microscopic and macroscopic appearances of the lungs, and I have plotted against the degree of oedema the systemic blood pressure and the amount of fluid transfused, mainly blood (Fig. II). In these cases, most of whom were suffering from varying degrees of toxaemia of pregnancy, the blood pressure is higher in the group with the greatest pulmonary oedema. This would be even more striking if we excluded two cases of chronic rheumatic carditis from the second group in whom the systemic blood pressure was normal, but a stenosed mitral valve upset the pulmonary circulation. In this group, therefore, it is more evident that the effect of the high systemic blood pressure on the left ventricle played a part in the production of pulmonary oedema. Maybe, also, the fluid administered to some of these patients accentuated the degree of oedema. However, other factors in toxaemia of pregnancy, not so easy to assess quantitatively, may also have affected the picture.

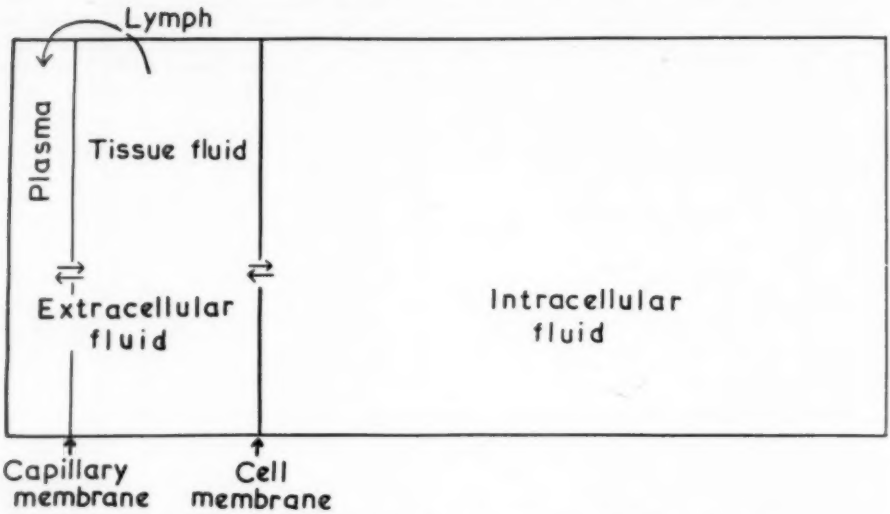

FIG. III. Diagrammatic representation of the fluid compartments of the body.

Because of this difficulty in assessing the causes and the development of pulmonary oedema in these clinical cases, we must turn to the laboratory experiment, both on man and animals, where we can study function in an endeavour to elucidate the fundamental principles involved. Then we must try to interpret our experimental results in terms of the clinical patient. Let us then see what controlled experiment has done to give us a clearer understanding of the changes in the fluid balance of the lungs that occur when the clinician observes that his patient is developing pulmonary oedema.

THE DISTRIBUTION OF FLUID IN THE BODY

It has been shown that within the organism water and the electrolytes are distributed throughout two main compartments, extra-cellular and intra-cellular. The intra-cellular water is on an average about 50 per cent. and the extra-cellular about 20 per cent. of the body weight (Fig. III). These two compartments are separated by the cell membrane through which there is a slow exchange of electrolytes, which cannot be explained simply by osmotic forces.

It is, however, the extra-cellular fluid in which we are mainly concerned, and the exchange that goes on through the capillary membrane between the two extra-cellular compartments, the circulating plasma on the one hand and the tissue fluid on the other. The composition of the extra-cellular fluid is almost identical, whether it is intra-vascular or extra-vascular, the chief difference being a higher protein content in the plasma than in the tissue fluid. The exchange between these two fluids depends almost entirely on the fact that the capillary wall is relatively impermeable to the plasma proteins, whereas water and electrolytes pass freely through this membrane. So, in the equilibrium that exists, the osmotic pressure of the proteins within the capillaries aided by the tissue tension counteract the capillary blood pressure. It is the balance of these pressures that is mainly responsible for maintaining the volumes of these two compartments.

Within the extra-vascular compartment of the extra-cellular fluid there is a specialized system, the lymphatics, whose main function in the normal organism is to return to the blood stream the large protein molecules and any large particles such as red cells that escape from the blood capillaries. Experiment has shown that the capillary membrane is not completely impermeable to the plasma proteins and that all or almost all the protein that becomes extra-vascular is re-absorbed into the blood stream by way

of the lymphatics (Courtice and Simmonds, 1949; Courtice and Steinbeck, 1950). A complete blockage of the lymphatics from a region leads to the local accumulation of the extra-vascular protein with resultant oedema (Drinker, Field and Homans, 1934). Normally, therefore, there is a slow extra-vascular circulation of plasma protein, varying in amount in the different tissues but amounting in all to between 50 and 100 per cent. of the total plasma protein per day, by which the tissue fluid is freed of protein, so preventing oedema (Courtice, 1952).

THE PULMONARY CIRCULATION

These general principles involved in the fluid balance of the body as a whole apply also to the lungs in particular. In considering the lesser circulation, however, we must remember that: (i) the only function of the lungs is the physico-chemical exchange of gases between the blood and air; (ii) all the tissue served by the lesser circulation has this same function and the whole cardiac output has to pass through this circulation. There is, therefore, little necessity for a complex regulating mechanism of the blood flow to different areas as there is in the greater circulation; (iii) there is also no necessity for a high rate of fluid exchange since the pulmonary capillaries are practically devoid of support, the glandular and muscular tissue of the bronchi being supplied by the systemic circulation; (iv) because of the close proximity of the pulmonary capillaries to the alveoli, any increase in the formation of the tissue fluid soon fills the alveoli, interfering with the function of the lungs.

The pulmonary artery after breaking up into smaller and smaller branches ends rather abruptly in a network of capillaries around the alveolar saccules. This capillary network is the richest in the body, the mesh being exceedingly small and often less than the diameter of the capillaries that bound it (Miller, 1937). In the systemic circulation the arterioles with their vasomotor control are of the utmost importance in the regulation of the volume of blood flowing to an organ or tissue depending upon its activity. In the pulmonary circulation there is less need for variations in arteriolar tone, since it is not so important to alter the distribution of the blood to the various parts of the normal lung. The pulmonary arterioles are supplied by the sympathetic nerves which are vasoconstrictor, but the balance of experimental evidence suggests that, as in the brain, these vasomotor nerves are not of great functional significance. In accordance with these anatomical differences in the two systems, the peripheral resistance to the ventricles, which mainly resides in the arterioles, is about ten times greater in the systemic than in the pulmonary circulation (Motley *et alii.*, 1947).

The pressures throughout the two systems confirm the low arteriolar resistance in the pulmonary arterioles. In the systemic system the pressure in the large arteries is at rest approximately 120 mm. of mercury in systole and 80 mm. of mercury in diastole. A large fall in pressure occurs in the arterioles with a resultant capillary pressure of 20-30 mm. of mercury. In the pulmonary circulation the pressures have been determined in animals (c.f. Wiggers, 1921; Hamilton, *et alii.*, 1939) and have in recent years been confirmed in man by the cardiac catheterization technique. Average figures obtained by some authors for man are given in Table 2. These experiments show a considerable drop of pressure from systole to diastole, indicating that there is a low peripheral resistance. There is also only a small fall from the mean pulmonary arterial pressure to the mean capillary pressure, compared with the very large fall in the systemic circulation. This again indicates that the arterioles do not play the same role in providing a peripheral resistance as they do in the systemic circulation.

Another interesting feature of the pulmonary circulation is the expansibility of the vascular bed. Normally in animals the lungs contain about 9 per cent. of the total blood volume during inspiration and about 6 per cent. during expiration (Wiggers, 1921). This may in certain conditions increase to about 20 per cent. of the total blood volume. In the normal animal this increase in volume of the pulmonary vascular bed may occur without any great increase in arterial or capillary pressure. It may be brought about by either an increased discharge of the right ventricle or an increased resistance in the systemic circulation, the "back pressure effect."

The effect of an increased discharge of the right ventricle can be observed in muscular exercise where the output of blood from the right side of the heart is suddenly increased. This is accompanied by an increased volume of blood in the lungs and an increased flow of blood through the lungs, but with no significant change in pressure. Average results obtained by two groups of workers are given in Table 3 (Riley *et alii.*, 1948; Hickam, and Cargill, 1948).

THE LYMPHATICS OF THE LUNG

To ensure the absence of fluid in the alveoli, any plasma protein that escapes from the circulating blood into the lung tissue fluid must be removed. This may be done in one of three ways — it may be coughed up, it may be digested by proteolytic enzymes and then absorbed into the blood stream or it may be absorbed by the lymphatics.

TABLE 2

The pressures in pulmonary artery and pulmonary "capillaries" in man at rest. mm. of mercury

Author	Pulmonary Artery			Pulmonary Capillary
	Systolic	Diastolic	Mean	
Doyle *et alii* (1951)	21	5	12	5
Dexter *et alii* (1950)	23	9	15	9
Riley *et alii* (1948)	20	9	13	—
Motley *et alii* (1947)	22	6	13	—

Increased resistance in the systemic circulation may be produced by adrenaline. The vasoconstriction causes a great increase in systemic peripheral resistance against which the left ventricle suddenly has to work. The output is decreased so that the pulmonary veins are distended and the pressure will tend to rise. This rise will be transmitted back through the pulmonary circulation. There results, therefore, in the normal animal an increased volume of the pulmonary vascular bed with a slightly elevated pressure throughout (Johnson *et alii.*, 1937).

Our conception, then, of the pulmonary circulation is one with a very high volume flow at a low pressure and against a low resistance, and in which the volume of the vascular bed is considerably expansile with but little dependence on vasomotor nerves. This expansibility means that the lungs can normally accommodate increased volumes of blood by dilatation of vessels and by the opening up of fresh capillaries without any great increase in pressure.

The lungs are provided with an abundant supply of lymphatics, a superficial set situated in the *pleura pulmonalis* and a deep set which accompanies the bronchi, pulmonary artery and its ramifications, and the pulmonary veins (Miller, 1937). The lymph flow is towards the hilum of the lung and then through the lymph nodes mainly to the right lymph duct (Warren and Drinker, 1942; Cameron and Courtice, 1946). In animals this duct can often be cannulated and lymph collected.

What we want to know from our lymphatic experiments is: (i) what part the lymphatics normally play in the fluid balance of the lungs, i.e., how much protein normally escapes from the pulmonary blood capillaries to be returned by the lymphatics and; (ii) the ability of these lymphatics to remove a considerable excess of extra-vascular protein such as occurs in pulmonary oedema from various causes.

I shall answer the first question now and leave the second until a little later. In experiments which I performed some years ago in conjunction with Professor G. R.

TABLE 3

The effect of mild exercise on pulmonary circulation in man (normal)

	Cardiac output l./min./m²	Pulmonary Artery Pressure mm. of mercury		
		Systolic	Diastolic	Mean
Riley *et al.* (1948). Mean figures for 3 subjects				
Rest	3.27	20	9	13
Exercise	5.40	23	4	11
Hickam and Cargill (1948). Mean figures for 8 subjects				
Rest	4.2	—	—	11
Exercise	6.1	—	—	13

Cameron, we collected the lymph from the right lymph duct of dogs. This lymph comes from the lungs and heart, and also, as we have since shown, from the pleura and diaphragmatic peritoneum. In Table 4 are shown average figures for the lymph flow and protein in the right lymph duct and these are compared with similar figures for the thoracic duct which drains mainly the abdominal viscera. It is evident that the amount of protein leaking from the lung capillaries is very much less than that leaking from the capillaries of the alimentary viscera. If we could collect the lymph coming only from the alveoli, i.e., only from the pulmonary circulation, the protein leakage would probably be much less than this table indicates. The lymph flow is also small, running parallel with the amount of extra-vascular protein to be removed. In these experiments we were struck by the very small lymph flow, sometimes practically none at all, from an organ through which such an enormous amount of blood is flowing, so that although the protein concentration in the lung lymph is high, the amount of protein escaping from the lung capillaries in a given time is very small, compared with that from the glandular tissue of the abdominal viscera.

Let me here digress for a moment to clarify our interpretation of these results in terms of capillary permeability. The percentage of protein in the lymph and, therefore, we think, in the tissue fluid, does not alone give any indication of the permeability of the capillary membrane to proteins. In equilibrium the level of protein in the tissue fluid in any one region is such that the resultant osmotic pressure of the plasma proteins is approximately balanced by the capillary pressure. If the capillary pressure is very low, non-protein fluid will be reabsorbed until the effective osmotic pressure balances this pressure, i.e., the extra-vascular concentration of protein will be high and approximate that in the plasma. With a higher capillary pressure, equilibrium will be reached at a higher effective osmotic pressure, i.e., the extra-vascular concentration of protein will be lower. The total amount of protein escaping from a region in a given time, however, does give an indication of the permeability of the capillary membrane, although the area of the capillary bed and the capillary pressure should also be considered.

It is evident that in the pulmonary circulation, very little protein escapes, but the capillary pressure is so low that non-protein fluid is reabsorbed to such an extent that the level of protein in the lymph is very high. In the abdominal viscera, on the other hand, not only is the level of protein high, but the actual amount of protein escaping in a given time is very high. It seems, therefore, as I have postulated elsewhere (Courtice, 1952), that in glandular tissue where the protein turnover is very

high, the capillary membrane is more permeable to proteins. Although the lungs are richly supplied with lymphatics, there is no necessity for a high extra-vascular circulation of protein in a tissue that has a low protein metabolism; so, we find that the capillary wall normally allows little escape of plasma protein from the large amount circulating through the lungs in unit time.

TABLE 4

The lymph flow in anaesthetized dogs. Average figures for a group of 21 animals for the right lymph duct and 4 animals for the thoracic duct

	Weight kg.	Lymph volume ml./hr.	Lymph protein g. per cent.	Lymph protein g./day
Right lymph duct	21	2.3	3.7	2.0
Thoracic duct	21	25.0	4.1	26.1

Average circulating plasma protein in each group = 55 g.

PRODUCTION OF PULMONARY OEDEMA

For fluid to accumulate in the lungs there must be at least one of the following factors present:

(i) Increased permeability of the capillary membrane to proteins,

(ii) Blockage of lymphatics,

(iii) Disturbance of the pressures which maintain the normal fluid exchange.

Increased permeability of the capillary membrane

Increased permeability of the capillary membrane to proteins may be brought about by various toxic agents, inhaled or injected, e.g., phosgene, lewisite, alpha-naphthyl thiourea (Cameron and Courtice, 1946; Cameron, Courtice and Short, 1947; Drinker, 1945; Richter, 1952). The lungs become grossly and diffusely filled with a protein-rich fluid resembling plasma in composition. This is brought about by an increased permeability of the capillary membrane without any significant alteration in the haemodynamics of the pulmonary circulation. It is remarkable how a change in the capillary membrane can give rise to such a massive outpouring of protein-rich fluid, even though the capillary pressure is so low. Once this membrane is damaged, however, there is no supporting tissue to resist the outgoing fluid and the alveoli become filled.

TABLE 5

The protein leakage from the circulating blood into the lungs in normal and phosgenized dogs. Average figures for a group of 21 normal animals and 8 animals after exposure to phosgene

	Weight kg.	Extra-vascular protein
Normal	21	0.08 g./hr.
Phosgenized	22	{ Lymph 0.2 g./hr. { Total 2.2 g./hr.

We now have to discuss the ability of the lung lymphatics to remove this excess of extra-vascular protein. In these conditions of acute pulmonary oedema, we have collected the lymph from animals and showed that the extra-vascular protein is greatly increased (Table 5). This table gives the mean escape of protein over the entire period from exposure to phosgene to death. The rate of protein loss from the capillaries is much greater than this just before death. The lymphatics are widely

dilated but show no evidence of blockage since the lymph flow is increased (Fig. IV). The lymph flow is only greatly increased when the oedema becomes extensive and parts of the lungs solid with fluid. The forceful expiration then causes a positive pressure in the thoracic cavity, forcing the lymph on.

problem further. Dr. Simmonds and I studied the removal of protein labelled with the blue dye, T1824, from the lungs of normal animals. We found this to be very slow compared with the lymphatic absorption of protein from the pleural or peritoneal cavities (Courtice, 1949), but our results suggested that apart from fibrin the

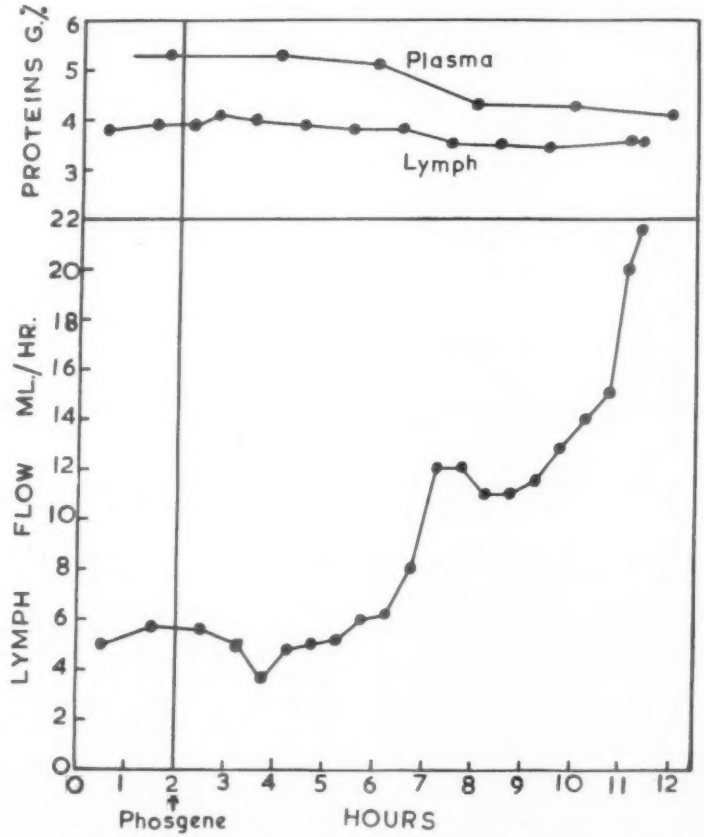

FIG. IV. The lymph flow from the right lymph duct of a dog before and after the onset of acute pulmonary oedema due to exposure to phosgene.

In contrast to the lymph flow in acute massive pulmonary oedema, the removal of the protein in the recovery phase is very slow and it usually takes about a week for the excess protein to be absorbed from the alveoli. This slow removal of protein from the alveoli in the recovery phase of pulmonary oedema led us to investigate the protein molecules are absorbed as such and are not first broken down by proteolytic enzymes (Courtice and Simmonds, 1949). Even though the protein was slowly absorbed from the lungs of a normal unanaesthetized animal, we found that anaesthesia and the recumbent posture reduced the rate of absorption to practically zero (Fig. V).

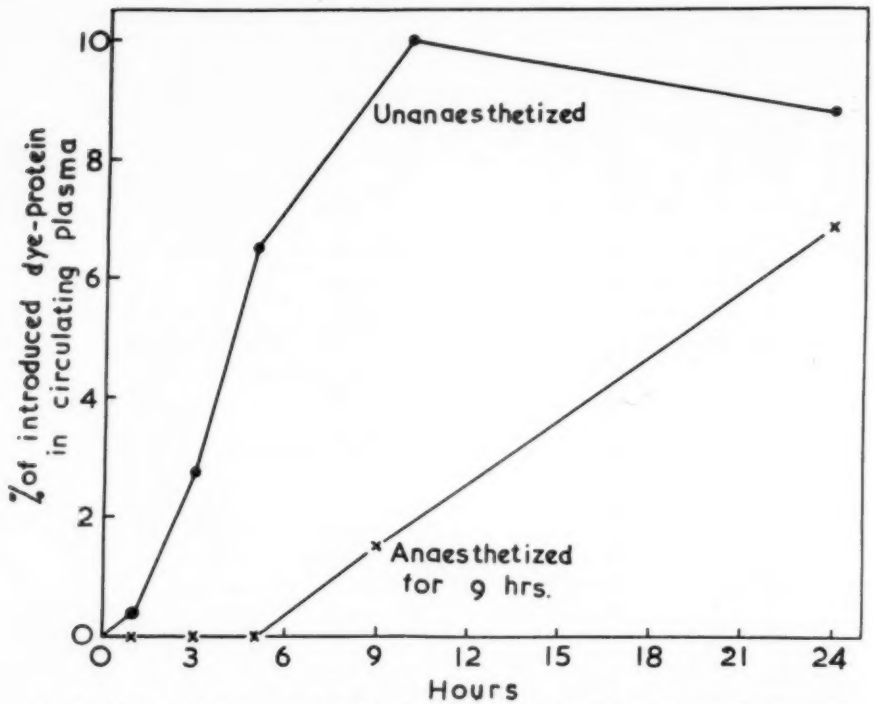

FIG. V. The effect of anaesthesia on the lymphatic absorption of plasma protein from the lung alveoli of rabbits after the introduction via the trachea of the animal's own plasma (1 ml./kg.) with the proteins labelled with the dye T1824.

Thus, in any type of exudate in the lungs due to a damage of the capillary membrane, whether by chemical toxins, bacteria or bacterial toxins, the extra-vascular protein will be high and the removal of this protein will be slow. The most rapid way of removing this fluid or exudate is by coughing it up. The ability of the patient to cough in the post-operative period may, therefore, be of considerable importance. Our experiments with anaesthesia also show in a conclusive manner the effect of movement of the lungs and of the bodily posture on the absorption of protein. From a post-operative viewpoint, movement and change of posture are of considerable significance in the prevention of an accumulation of protein and of fluid in the alveoli.

Disturbance of the balance of pressures

The pressures involved are the capillary pressure aided by the negative intra-thoracic pressure and the opposing osmotic pressure of the plasma proteins. The most significant changes in these pressures which lead to oedema probably occur in the capillary pressure. We have seen that in the normal individual the expansibility of the pulmonary vascular bed prevents any appreciable rise in the capillary pressure. In certain circumstances, however, the capillary pressure may be considerably increased.

(i) Diseases of the heart and lungs. Diseases of the heart and lungs may impair the expansibility of the pulmonary bed or reduce the reserve in this expansibility, so that when an extra amount of blood is thrown in, the pulmonary blood volume will only be increased by a greatly increased pressure. Experiments have been performed which support this view. For example, whereas exercise in the normal individual gives rise to very little change in the pulmonary pressures, this is not so in patients with certain lung or heart disease. Average results from two groups of investigators are given in Table 6. These results can be

explained, in part at least, by a narrowing of the pulmonary vascular bed in such chronic pulmonary diseases as emphysema and to an already expanded vascular bed in the chronic cardiac conditions.

TABLE 6

The effect of exercise on mean pulmonary artery pressure in man

	Mean pulmonary artery pressure mm. of mercury	
	Rest	Exercise
Riley *et alii* (1948)		
Normals (3)	13	11
Chronic pulmonary disease (8)	20	34
Hickam and Cargill (1948)		
Normals (8)	11	13
Congestive Cardiacs (8)	36	54
Mitral Stenosis (4)	24	39
Pulmonary Emphysema (4)	24	37

It is evident that in these conditions of the heart and lung, the pulmonary capillary pressure may be raised, perhaps insufficiently to cause a significant increase in the extravascular fluid of the lungs. Since the osmotic pressure of the plasma proteins is normally much greater than the capillary pressure, this reserve in the balance of pressures has to be overcome before there will be any appreciable increase in the volume of tissue fluid. If, however, the output of the right ventricle is suddenly increased, as in muscular exercise, or if for some reason there is a general vasoconstriction of the systemic circulation, as sometimes occurs suddenly, the pulmonary vascular bed will be called on to accommodate a greater volume of blood. If the limit of expansibility has already been reached, this extra burden increases the pulmonary pressures and acute oedema may result.

(ii) Gravity. If an anaesthetized animal is kept in one posture for many hours, the dependent parts of the lung will become grossly oedematous. Owing to gravity the blood will tend to pool in these dependent parts and the capillary pressure will increase because of the force of gravity. The extravascular fluid will accumulate and the lymphatic drainage practically ceases because of insufficient movement. In this way oedema also forms in debilitated patients after operation. The importance of change of posture in the prevention of this type of oedema is evident.

(iii) Transfusions. Another important factor in the post-operative treatment of patients which may affect the pulmonary pressures is the intra-venous transfusion of various fluids. Dr. P. Korner and I are at present investigating this problem with the object of finding what factor or factors will precipitate pulmonary oedema.

In these experiments we have used rabbits and have transfused them with a constant drip into the ear vein with Ringer-Locke solution over a period of five hours. At the end of this time we have assessed at post-mortem the degree of pulmonary oedema, graded 0, 1, 2 and 3. This has been checked by measuring the lung/body weight ratio, and it is evident that the assessment runs parellel with this ratio (Fig. VI).

In experiments on normal, unanaesthetized rabbits we found, as others have done, that large volumes of fluid may be transfused before pulmonary oedema is produced. With increasing rates of transfusion the urine

flow increases very greatly, but the plasma volume and the extra-cellular fluid also increase. In Fig. VII is plotted the increased tissue fluid load (obtained by subtracting the volume of urine and the excess fluid in the plasma from the amount transfused) against the degree of pulmonary oedema. In Table 7 actual figures are given for the increase in the tissue fluid in the systemic circulation and in the pulmonary circulation, observed in typical experiments.

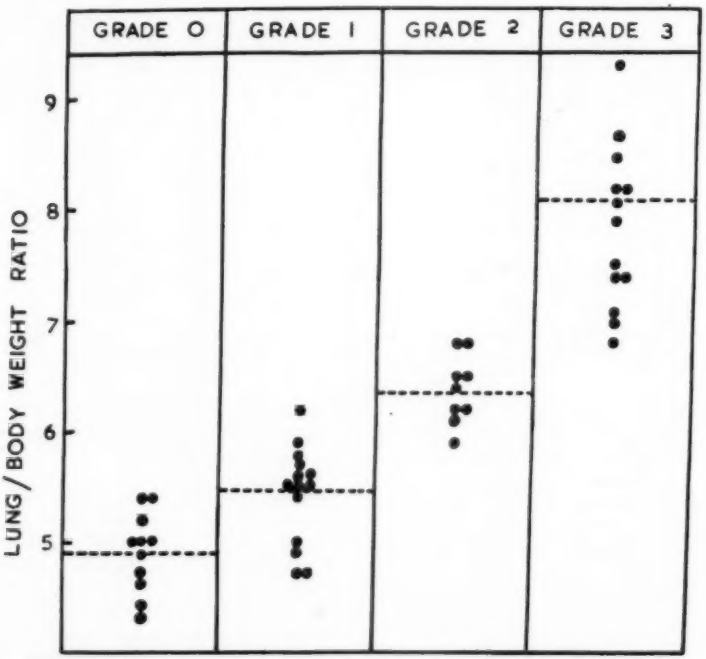

FIG. VI. The relation between the assessed grade of pulmonary oedema and the lung/body weight ratio in rabbits.

It is evident that a great deal of fluid can be transfused into normal animals, but most of this passes out into the systemic tissues and not into the lungs. The low capillary pressure and the expansibility of the pulmonary vascular bed ensure that very little filtration occurs in the lungs whereas large quantities are filtered in the renal glomeruli where the capillary pressure is very high and also in the systemic tissues generally where the capillary pressure is much higher than in the lungs. By increasing the load sufficiently, however, the pressure in the pulmonary capillaries rises sufficiently to cause a constant increment in pulmonary oedema for each increment in the systemic tissue fluid. Nevertheless, it is remarkable to observe how the adaptations of the pulmonary circulation to a rising blood volume ensure that only a minimum of fluid escapes into the alveoli. In these experiments the amount of urine increased very greatly with increasing transfusion rates. If, therefore, the kidneys are not functioning or if they are not functioning normally, e.g., in certain conditions of sodium retention, the amount of fluid transfused to produce a given overloading of the extra-cellular spaces will be much less.

We have as yet measured the pressures in only one animal and found that the left atrial pressure rises gradually with transfusion. This pressure would reflect the pulmonary capillary pressure. In man the pulmonary pressures have been determined before, during and after the rapid transfusion of 1000 c.c. in 10 minutes (Doyle *et alii*.,

1951). The transfusion and resultant increase in blood volume cause a rise in pulmonary artery and pulmonary "capillary" pressures with a rapid fall soon after the end of the transfusion and with no signs of pulmonary oedema. The rise in pulmonary capillary pressure was, however, from the normal level of 5 mm. of mercury to a maximum of only 12 mm. of mercury which is still very much less than the osmotic pressure of the plasma proteins. These experiments support the conception of an expansile pulmonary vascular bed in which there is normally a considerable reserve in the mechanisms protecting the alveoli from excess fluid.

lymph flow from the right lymph duct. We have used degrees of anoxia similar to those used by Drinker. Our findings are shown in Fig. VII. In these experiments we transfused unanaesthetized rabbits as before, but instead of breathing air (20.93 per cent. oxygen) they breathed various mixtures deficient in oxygen. These points are evident from these experiments

(a) Anoxia alone for five hours does not produce any excess tissue fluid in the lungs.

(b) When the circulation is overloaded by transfusion, anoxia precipitates oedema at a lower loading than when air is breathed.

(c) Once an excess of pulmonary tissue fluid arises, the increase in this runs parallel with the increase in the systemic tissue fluid. This is similar to the observations made when air is breathed.

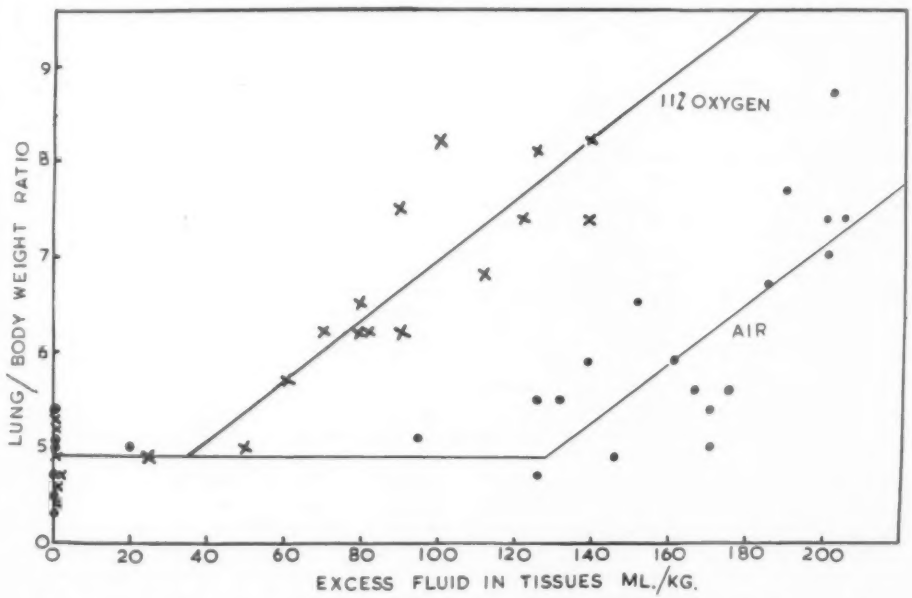

FIG. VII. The relation between the excess tissue fluid in the systemic tissues and the degrees of pulmonary oedema, determined by the lung/body weight ratio, following transfusions of Ringer-Locke solution for 5 hours at varying rates.

• Experiments in which the animals breathed room air.
X Experiments in which the animals breathed 11 per cent. oxygen.

(iv) Anoxia. We are now using these initial experiments as a baseline to ascertain whether any factor may precipitate oedema. The first factor which we have investigated is anoxia. Drinker has maintained that oxygen lack increases the permeability of the pulmonary capillaries, his views being based in large part on experiments concerning the

TABLE 7

The increase in the tissue fluid of the tissues supplied by the systemic circulation and of the lungs supplied by the pulmonary circulation as a result of increasing transfusions of Ringer-Locke solution in rabbits over a period of five hours

Air breathed	Systemic circulation ml./kg.	Pulmonary circulation ml./kg.
Air	127	0
"	174	0.6
"	200	3.0
11 per cent. oxygen	60	0.3
"	85	1.2
"	131	2.8

The fact that fairly severe anoxia alone (10.5-11 per cent. oxygen) does not cause any measurable increase in the amount of pulmonary tissue fluid, indicates that the permeability of the capillaries to protein cannot be altered. If the permeability were appreciably altered, protein would accumulate in the alveoli because of the slow lymphatic removal.

The onset of pulmonary oedema in anoxia with lower transfusion rates again does not suggest any alteration in the permeability of the capillary membrane. If this altered, the increase in pulmonary oedema with increasing transfusions would not run parallel with a similar increase when air is breathed. It seems, therefore, that in these experiments the onset and development of pulmonary oedema can be explained merely by changes in pressures.

A study of the cardiac output in these unanaesthetized animals has been made using the direct Fick method, samples of blood being obtained from the right heart and from an artery by fine polythene cannulae. Breathing 11 per cent. oxygen depresses the myocardium leading to a sudden fall in stroke volume and cardiac output. The systemic blood pressure momentarily falls but soon rises again as a result of the reflex constriction of the systemic arterioles (Fig. VIII). The systemic system thus decreases in volume, so an additional strain is thrown on the systemic veins and on the expansile pulmonary vascular bed. The cardiac output goes on falling during the five hours, but vasoconstriction prevents a great fall in the blood pressure.

We are postulating, therefore, that in the anoxic, non-transfused animal the expansibility of the pulmonary vascular bed allows an increase in the volume of pulmonary blood without an appreciable increase in capillary pressure. When the load of transfusion is added by increasing the blood volume the already enlarged volume of pulmonary blood is further increased so that an excess pulmonary fluid will result from a lower transfusion load. Once this limit is reached then an increase in the load will produce the same increase in pulmonary oedema, whether the animal is anoxic or not.

These experiments perhaps help to explain why a patient might be progressing well post-operatively and then suddenly develop pulmonary oedema. Some extraneous factor, for example emotion or pain, may cause a sudden intense vasoconstriction of the systemic vessels with a resultant increase in the resistance to the left ventricle and an increased expansion of the pulmonary circulation. If the lungs are normal and the circulation is not overloaded or even if the lungs and heart are not normal but the circulation is not over-loaded, the pulmonary vascular bed will probably be able to cope with the increased pulmonary blood volume without a considerable rise in pulmonary capillary pressure. If, however, the patient has been over-transfused, but not sufficiently to cause pulmonary oedema in itself, this extra factor will raise the capillary pressure above the safety limit and oedema will result.

So we see how it is that despite the rigours of the external environment to which the alveoli are always exposed and despite the added internal stresses sometimes imposed in this modern age by a too ardent transfuser, the transfer of oxygen and carbon dioxide across the capillary membrane is rarely affected. Sometimes, however, these extraneous forces are too great and fluid fills the alveoli instead of air. Even though pulmonary oedema may not be a major hazard for the surgeon, these experiments

will perhaps help us towards a fuller understanding of the fascinating way the alveoli are protected and allowed to carry on their physico-chemical function.

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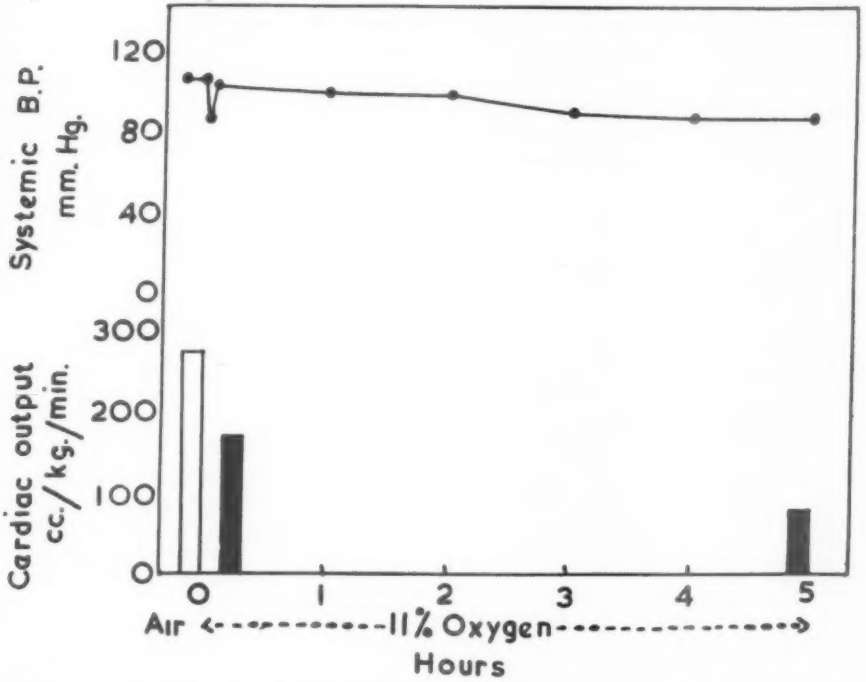

FIG. VIII. The effect of anoxia (11 per cent. oxygen) on the blood pressure and cardiac output of unanaesthetised rabbits. Similar changes were observed with anoxia in the transfused animals.

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HIATUS HERNIA IN INFANTS AND CHILDREN

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Royal Alexandra Hospital for Children, Sydney

THE object of the present report is to draw attention to the occurrence of hiatus hernia in infants and young children. If this condition is treated at an early stage the occurrence of serious complications may be prevented. Up to the present time there has not been a great number of these cases reported in children and the problem of hernia through the diaphragm as far as children are concerned has been thought to be mainly the problem of hernia through the foramen of Bokdalek, usually on the left side. In adults on the contrary the great majority of cases of diaphragmatic hernia occurs at the oesophageal hiatus. These facts may be illustrated by the figures from the Mayo Clinic (Harrington, 1945) and the Babies Hospital, New York (Donovan, 1945).

Reference to Table 1 will show that in Harrington's series, 231 occurred at the oesophageal hiatus and 33 were described as being of the short oesophagus type, but all these cases were seen in adult life. In the same journal Donovan reported 17 cases from the Babies Hospital. Although he states that "the most common congenital defects in the diaphragm are in order of their occurrence: (1) oesophageal hiatus; (2) foramen of Bokdalek; (3) foramen of Morgagni; (4) defects in the "dome;" there was only one case in his series occurring at the oesophageal opening and this could perhaps be classed as a para-oesophageal hernia as at operation "the entire stomach, the first and second parts of the duodenum, and almost all of the transverse colon were found to be in the chest, having herniated through a large esophageal hiatus."

The 10 cases occurring at the Royal Alexandra Hospital for Children were taken from the hospital records of the last ten years, and include the 4 cases described in this report which are the first cases of this type reported from this hospital. The figure (?7) implies that there have probably been 5 additional cases which have not been recognized as suffering from hiatus hernia.

More recently Husfeldt (1951) has reported a series of 24 cases in children with a sliding type of hernia at the oesophageal hiatus and divides them into two types. In the first type the oesophagus is shorter than normal and opens directly into the hiatal hernia. There is a constriction usually at the level of the seventh or eighth dorsal vertebra indicating the cardiac orifice and the part of the stomach passing through the hiatus is broad showing that the hiatus is larger than normal. Regurgitation is demonstrated by fluoroscopy. In the second type, which it is suggested may be the precursor of the first, the oesophagus is dilated and there is no constriction at the cardia, but the oesophagus is directly continuous with the fundus giving the characteristic funnel-shaped appearance in the X-ray film. Radiographic examination will also demonstrate continuation of the gastric mucosa into the hernia in both types.

Oesophagitis and ulceration

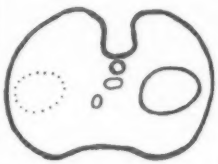
The result of incompetence of the cardiac sphincter is that reflux of the acid gastric contents occurs, oesophagitis develops and ulceration may occur at the lower end of the oesophagus. This will frequently be followed by cicatricial changes with shortening and fixing of the oesophagus and it is this fact that makes it important to recognize these cases in early life so that surgical repair may be undertaken before the oesophagus "freezes" in its retracted position (Allison, 1951).

Symptoms and Diagnosis

The outstanding symptom in infancy is vomiting which may occur early enough to suggest the diagnosis of pyloric stenosis and

TABLE 1

THE INCIDENCE OF VARIOUS FORMS OF DIAPHRAGMATIC HERNIA

	Varieties	Harrington	Donovan	Children's Hospital
	Morgagni	8	2	1
	Bokdalek	9	12	5
	Para	72	1	0
	Hiatus	198 33 c.s.o.	0	2
	Cicatrization	0	0	2 (possibly 7)
	Eventration	0	1	0
	Congenital Absence	12	1	0
	Traumatic	72	0	0

The figures for Harrington's and Donovan's series represent the position in 1945. c.s.o. refers to the 33 cases of congenital short oesophagus included by Harrington in the cases of hiatus hernia. The last column represents the cases of diaphragmatic hernia operated on at the Royal Alexandra Hospital for Children during the 10 year period 1941-1951. Under the heading cicatrization are included two cases reported in this article where cicatrization with oesophageal obstruction was the outstanding feature. It is considered that during this period there were possibly 5 other cases causing oesophageal obstruction that were not recognized as suffering from hiatus hernia.

some cases have been operated on in the belief that this condition was present. The vomitus may be blood-stained and the stools may be tarry, and these signs are evident when oesophagitis or ulceration have developed. Older children may complain of dysphagia and pain during meals at the lower end of the sternum.

CASE REPORTS

I have been fortunate in seeing 4 cases in a relatively short period which illustrate many of the points mentioned above.

FIG. I. Case 1. Stenosis of the lower end of the oesophagus with gastric rugae extending above the diaphragm.

Case 1. Mr. M. Sofer Schrieber's case.

This boy of seven years was admitted for investigation on 24th July, 1951, with a history of inability to swallow solid foods since the age of twelve months. He was a mental defective, unable to talk, and his arresting physical condition of underdevelopment and dehydration was explained by the fact that he had lived for most of his life on fluids only and that he vomited almost immediately after taking any food. X-ray examination showed a narrowing of the oesophageal lumen and tortuosity of the lower end which was regarded as being due to a stricture of unknown causation. The oesophagus was gradually dilated at weekly intervals and, as dilatation proceeded, he began to take food and improve in physical condition. Further X-ray examination was undertaken to determine whether a hiatus hernia was present, but this could not be demonstrated by screening although the films suggested that gastric mucosal folds extended above the hiatal opening (Fig. I). In November, 1951, as a result of his dilatations, a remarkable change had occurred in his condition. He was rapidly putting on weight, could take all solid foods without vomiting, and had improved mentally, displaying an alert interest in the ward activities, and was even making attempts to talk. An exploratory operation was considered justifiable to determine the condition of the hiatal opening in view of the suggestive X-ray examination.

FIG. II. Case 1. Barium swallow taken after the oesophagus had been dilated and the hernia repaired.

The operation was performed by Mr. Sofer Schreiber, and the oesophagus was exposed through a left thoracotomy. By separating the dense inflammatory adhesions in which it was buried the oesophagus was freed as high as the inferior pulmonary vein and its lower end followed down to the hiatus which was obviously dilated. The diaphragm was incised an inch from the hiatus in a radial direction and the hernial sac identified from below. This was incised and the oesophagus brought down into the abdominal cavity and fixed in its new position by suturing the phreno-oesophageal ligament to the lower surface of the diaphragm as described by Allison. The crura were repaired behind the oesophagus by silk sutures, and the

diaphragm sutured and the chest closed. Subsequent X-ray examination showed that the herniated oesophagus had been satisfactorily reduced. His progress has continued to be satisfactory but he will be re-admitted for further dilatation at a later date (Fig. II).

Case 2. The second case was referred to me by Professor Dods.

This was a boy of 6 years, the victim of cerebral palsy manifested by quadriplegia with athetosis. He was an intelligent boy and was responding well to his management at the Spastic Centre. For a period of eighteen months before his admission he had had haematemesis on several occasions. Some of these episodes were of minor character, but on one occasion there had been a massive haemorrhage necessitating transfusion. Examination by barium meal after this attack had shown a dilated stomach and delayed emptying time, but on this occasion the oesophagus was not examined. Following a further massive haemorrhage he was admitted to the Institute of Child Health. Investigation by barium swallow showed a small hiatus hernia involving the cardiac end of the stomach and oesophagoscopy showed no obstruction. Operation was undertaken and the hernia displayed. This was repaired following the same technique as that described in the first case. His subsequent progress has been satisfactory and there has been no further haemorrhage.

The first case illustrates the effects of cicatricial narrowing of the oesophagus following ulceration with vomiting as the predominant symptom, and in the second case haemorrhage was the outstanding symptom drawing attention to the condition before stricture had occurred.

Case 3. This case was also referred by Professor Dods.

A child aged 6 months on admission, was well until the age of two months when she vomited dark brown fluid (presumably blood). She continued vomiting in attacks with intervals of freedom lasting one or two weeks. Two weeks before admission the vomiting had become more frequent and it was dark brown on occasions. She was anaemic and losing weight. Examination by fluoroscopy showed "no delay in the passage of the bolus, but in the Trendelenburg position there appears to be a small hiatus hernia of the cardiac end of the stomach." The importance of the examination in the recumbent position is stressed by all writers and it may be necessary to employ pressure on the abdominal wall to demonstrate the reflux into the oesophagus. In this case, subsequent X-ray examination in the inverted position showed "a marked bulge at the lower end of the oesophagus the appearance suggesting a congenital short oesophagus with partial thoracic stomach" (Fig. III).

Operation was undertaken and the defect displayed. There was no difficulty in bringing the stomach down below the diaphragm, so that the suggestion of a congenital short oesophagus was not confirmed.

FIG. III. Case 3. Hiatus hernia diagnosed as congenital short oesophagus with thoracic stomach but easily reduced below the diaphragm at operation. Rays taken in the inverted position.

Case 4. This case is included by courtesy of Mr. E. S. Stuckey.

A baby of 9 months was admitted with the history that at the age of two months he had vomited dark blood with mucus and continued to do so every few days, the vomitus containing blood sometimes bright and sometimes dark. During the month preceding admission he had lost one pound in weight and the motions were frequently tarry in character. He was anaemic and was given a transfusion on admission. Routine examination failed to show a definite cause for the bleeding. A barium swallow showed "fairly constant dilatation at the lower end of the oesophagus present in all positions on occasions. This is probably a phrenic ampulla but could possibly be a small hiatus hernia of short oesophagus type" (Fig. IV).

The child was allowed to go to the follow-up department for further observation and six months later was re-admitted. There had been no further vomiting or melaena but there was frequent regurgitation of mucus before feeding. At this time oesophagoscopy was performed and "the impression

FIG. IV. Case 4. Hiatus hernia before operation.

FIG. V. Case 4. After operation with successful reduction

was gained that the oesophagus was abnormally short." An obstruction was encountered high up and the instrument would not pass beyond this point.

X-ray examination again suggested a small hiatus hernia of the short oesophagus type.

Operation was performed and a sliding hernia was demonstrated with a definite sac. Repair was effected in a similar manner to that employed in the other cases and subsequent X-ray examination demonstrated the cure. The oesophagus was not abnormally short and could be brought below the diaphragm without difficulty (Fig. V).

DISCUSSION

These last two cases represent the satisfactory results that may be expected to follow early diagnosis and correction of the defect before cicatrization and shortening of the oesophagus has occurred. Not all of the cases require immediate operation and a waiting policy may be justified in some, but when haemorrhage becomes a prominent feature the indications for operation are clear. These cases serve to illustrate the necessity for careful X-ray examination in babies with unexplained vomiting attacks and the importance of examination in the Trendelenburg position without which it may not be possible to satisfactorily demonstrate the defect.

The question may also be raised as to whether such a condition as congenitally short oesophagus with thoracic stomach does in fact exist or whether cases described as suffering from this condition have in reality a sliding hernia at the oesophageal hiatus. Husfeldt (1951) doubts its existence or else considers that it must be extremely rare.

There are difficulties in diagnosis as the lower end of the oesophagus may be lined by gastric mucosa. Also heterotopic gastric mucosa may be found in the oesophagus as in other parts of the alimentary tract. This is commonest in the post-cricoid region and islets varying in size from minute areas to those having a diameter of from two to three centimetres have been found. Ulceration may be found when these islets are in the lower third and cicatrization and stenosis will follow. Bosher has reported a case of this type in a man of sixty years in whom resection of the lower half of the oesophagus was necessary for a stricture commencing at the arch of the aorta. In contrast to this

extreme organic change Lawler and McCreath (1951) have drawn attention to the occurrence of regurgitation without hiatus hernia in many cases of dyspepsia in adults which have been thought to have no organic basis and which can be relieved by maintaining the upright posture for some hours after taking food. Presumably this condition may occur in children, and this fact emphasizes the importance of displaying the defect by accurate radiological studies before the diagnosis of hiatus hernia is made.

A consideration of the cases reported here has reminded me that in a previous issue of this Journal, Hipsley (1942) reported six cases of oesophageal obstruction in children and his conclusion was that "the underlying cause of these cases was a congenital narrowing due to imperfect development of a section of the oesophagus similar to the narrowing sometimes found in other portions of the alimentary canal." It is impossible to deny that this may be one cause of oesophageal obstruction and, in fact, one of the cases reported was found to have a diaphragm across the lower end of the oesophagus with an opening two millimetres wide near its centre. This was obviously a developmental defect. But in most of the other cases the meticulous case reports and X-ray illustrations enable one to hazard a guess that some at least of these cases were suffering from hiatus hernia. One child had been operated on for pyloric stenosis at the age of twelve days because of the forceful character of the vomiting, but no pyloric obstruction was found. This child later continued to vomit and suffered from melena, and at the age of four years oesophageal obstruction was demonstrated, the oesophageal walls being rigid and resistant to dilatation, suggesting the condition found in Case 1 of this report.

In the records of the Children's Hospital each year a number of cases are recorded with the diagnosis of oesophageal stricture, and I feel that many of these cases may, with further investigation, be shown to be suffering from hiatus hernia.

SUMMARY

1. Four cases of hiatus hernia are reported in children. These have been verified by operation.

2. The symptoms and possible sequelae of this condition are outlined.

3. Attention is drawn to the importance of this condition as a cause of stenosis of the lower end of the oesophagus.

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THE URETER AFTER RADICAL HYSTERECTOMY

A PRELIMINARY REPORT

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WERE it not for the presence of the ureters the radical operation for carcinoma of the *cervix uteri* would be little more than a technical exercise in destructive surgery comparable with procedures such as radical mastectomy.

Since, however, the principles involved demand the widest possible excision of the pelvic cellular tissues with the uterus and *adnexa*, much of the vagina and the pelvic lymph nodes, it means that a considerable portion of each ureter must be freed from its surroundings. This entails the immediate risk of necrosis from vascular deprivation as well as the possibility of late involvement of the ureter in post-operative scar tissue.

TABLE 1

GROUP A. (16 cases)
NO ABNORMALITY IN POST-OPERATIVE PYELOGRAM.
15 Alive and well.
1 Alive with recurrence.

GROUP B. (3 cases)
SLIGHT CHANGES IN POST-OPERATIVE PYELOGRAM.
SYMPTOMLESS AND NON-PROGRESSIVE.
3 Alive and well.

GROUP C. (5 cases)
PROGRESSIVE URETERIC OBSTRUCTION.
(i) 3 due to recurrent growth. 2 Alive: 1 Dead.
(ii) 2 due to effects of operation. 1 Alive: 1 Dead.

GROUP D. (9 cases)
URETERO-VAGINAL FISTULA.
(i) UNILATERAL (5 cases). ALL CLOSED SPONTANEOUSLY.
1 Died recurrent growth.
4 Alive — all with gross renal damage.
(ii) BILATERAL (4 cases).
1 REFUSED TREATMENT.
Persistent fistula with gross renal damage.
3 TREATED BY URETERO-SIGMOID ANASTOMOSIS.
1 Died post-operatively.
1 Died 1 year after operation from recurrence.
1 Alive and well 15 months after operation.

Although their incidence may be lowered by improvement in technique, these complications will continue to occur in some proportion and should be regarded, to a certain

extent, as part of the price which must be paid if the attack is to be truly radical and if surgery is offered to other than the more favourable cases.

It has been pointed out by Stanley Way (1951) that the ultimate fate of the ureters following radical hysterectomy is not known, since no studies appear to have been made on such cases. It may be of interest, therefore, to present frankly our difficulties and disasters in the Radio-Surgical Clinic of the Women's Hospital, and to put forward certain views which we are beginning to formulate regarding these problems.

FIG. I: I.C. Stage 1 carcinoma cervix. Pre-operative pyelogram. No abnormality detected.

CLINICAL MATERIAL

Of 70 cases operated on since the beginning of 1950 there are 33 in whom pre- and post-operative pyelography was performed. These may be divided into four groups (Table 1):

(a) Sixteen cases in which no abnormality was demonstrated either before or after operation.

(b) Three cases in which slight dilatation of the renal pelvis and ureter was demonstrated post-operatively on one or both sides. In these the condition has been non-progressive and symptomless and they have remained clinically well.

(c) Five cases in which the kidneys have progressively deteriorated. Two of these cases represent the effects of recurrent growth while a third is probably of the same nature. The other two are due to ureteric stricture without evidence of growth.

(d) Nine cases of uretero-vaginal fistula. Of the problems arising from this material two will be discussed — non-malignant ureteric obstruction and uretero-vaginal fistula.

FIG. II : I.C. Fourteen months after operation. Tumour due to right-sided hydronephrosis now palpable. Right nephrectomy performed shortly afterwards.

Non-Malignant Ureteric Obstruction

Two of the cases in Group (C) are clearly complications of the operation. They show the need for careful post-operative observation and for an early decision to take active steps to preserve renal function before irreparable damage has occurred.

One patient died eight months after operation from pyonephrosis having, in the *interim*, suffered several attacks of pyelonephritis and having shown in successive pyelograms evidence of ureteric obstruction. Her pre-operative pyelogram was normal and no residual growth was found at autopsy.

The other patient, who was operated on nineteen months ago, has recently undergone nephrectomy for gross hydronephrosis. Her remaining kidney and ureter showed considerable dilatation and her prognosis is poor. To date there is no clinical evidence of recurrence of the growth. Her pre-operative pyelogram showed no abnormality (Figs: I and II).

FIG. III : J.F. Stage 1 carcinoma cervix. Pre-operative I.V.P. No abnormality detected.

It is evident that in both these cases our inactivity has been disastrous. It may be stated that in both instances radical surgery was preceded by treatment with full radon dosage and deep X-ray therapy, but it might be unwise to draw conclusions too hastily from this. The fact is that from time to time surgery, which was not at first envisaged, may have to be undertaken after such treatment. Moreover, with constantly improving radio-therapeutic technique, undesirable side-effects are progressively diminishing.

Far more significant, in my opinion, is that both these patients showed evidence of renal infection. Perhaps we were lulled into a false sense of security by a fairly rapid response to chemotherapy. Perhaps we were too easily satisfied that infection was eradicated, and were not sufficiently aware of the implication that such attacks are almost certain to be due to ureteric obstruction. At all events, we now feel that an attack of pyelonephritis is an indication for careful, frequent and sustained observation and review.

During the past eighteen months we have performed uretero-sigmoid or uretero-caecal anastomosis in 20 instances, mainly for palliation, and the experience thus gained has led us to believe that in both the cases just discussed, we should have employed this procedure as soon as ureteric obstruction was apparent.

FIG. IV : J.F. Six weeks after radical hysterectomy which was followed by development of uretero-vaginal fisula. Both kidneys excrete dye in normal time. Bilateral hydronephrosis and hydro-ureter. Obstructions appear to lie at pelvic brim. Right lower ureter is of normal width. Left lower ureter less well-defined and irregular—fistula may be from this side.

Uretero-vaginal fistula

In consequence of the radical nature of the operation, and because our material includes a number of late recurrences after radiotherapy, the incidence of this complication has been extremely high and we have been faced with problems of prevention and treatment.

To date, 9 uretero-vaginal fistulae have occurred in 70 operations—a rate of 13 per cent. Four followed surgery for late recurrence after previous radiotherapy, and clinically were Stage 2 in one instance, and Stage 3 in three. One fistula occurred after

radical surgery for a stump adenocarcinoma with parametrial involvement. Of the remaining 4 cases, 3 were clinically Stage 2, but of these 2 had node metastases, whilst one was Stage 1.

FIG. V : J.F. One year after operation. Right hydronephrosis perhaps a little less. Left kidney functionless. No discharge from fistula for two months.

FIG. VI : A.H. Stage 3. Recurrent carcinoma cervix. Pre-operative I.V.P. showing slight dilatation of left renal pelvis and stasis of dye in left ureter below the pelvic brim. Concentration of dye in right pelvi-calycean system is below the normal density.

In 5 instances there was a unilateral uretero-vaginal fistula, while four times bilateral fistulae were present. Of the 5 unilateral fistulae all became dry spontaneously, but their subsequent history gives no cause for complacency.

One patient has died with progressive bilateral ureteric obstruction due to gross pelvic recurrence. The other 4 show no evidence of growth, but 2 have a functionless kidney on the side of the closed fistula with contra-lateral hydronephrosis (Figs. III, IV and V). One has bilateral hydronephrosis, and one has gross unilateral hydronephrosis on the side corresponding to the fistula.

FIG. VII : A.H. I.V.P. Four weeks after radical hysterectomy, during which left ureter was ligated below the pelvic brim and the lower ureter was excised with the growth. This was followed by development of a uretero-cutaneous fistula. Pyelogram shows fairly gross right hydronephrosis and hydro-ureter with some dilatation on the left side.

The 4 bilateral fistulae have progressed as follows: One patient refused treatment and remains wet, with rapidly increasing bilateral hydronephrosis, punctuated by attacks of pyelonephritis.

FIG. VIII : A.H. I.V.P. Four months after original operation and eight weeks after left uretero-sigmoid anastomosis and division of a band pressing on the right ureter. Three weeks before this film was taken an extravasation of urine from the right ureter was drained and a second uretero-cutaneous fistula established. Increase in the dilatation of the left renal pelvis is noted. There is further dilatation on the right side, and the dye-filled fistulous cavity is seen below the pelvic brim on the right side.

Three cases underwent uretero-sigmoid or caecal anastomosis. Of these one died post-operatively as a result of complications following a burst abdomen. One showed significant improvement both clinically and in the pyelographic findings, and she led a comfortable life until growth recurrence ended the story (Figs. VI, VII, VIII and IX). One patient has done well, X-ray appearances showing a satisfactory state of affairs which has been paralleled by a smooth clinical course devoid of both infective episodes and physiological upsets. She leads an active and happy life (Figs. X, XI, XII and XIII).

DISCUSSION

The story of these fistulae is one of avascular necrosis of the ureter with a retro-peritoneal extravasation of urine and fistula formation. The consequences, which could be predicted from elementary pathological principles, are the occurrence of ureteric stricture, followed by hydro-ureter and hydronephrosis, with almost inevitably recurrent infective episodes, and finally irreparable damage to the kidney. It is this process which results in the so-called closure of many fistulae, and it is strikingly borne out in our material.

FIG. IX : A.H. I.V.P. Five months after left uretero-sigmoid anastomosis and freeing of right ureter, showing marked improvement on both sides. The fistula on the right side had drained freely and the changes following the radical hysterectomy six months previously had had time to subside. Note the extensive fistulous cavity in the pelvis. Right uretero-caecal anastomosis was performed shortly afterwards.

The implication which we draw from the foregoing is that spontaneous closure of a uretero-vaginal fistula is far from being a desirable end. The patient is forced to undergo an uncertain and often prolonged period of discomfort, with no guarantee that she will ultimately be relieved without operation. While awaiting death of the kidney, she is almost certain to undergo urinary tract infections, which not only produce ill-health but place the other kidney at risk. And, finally, with the ever-present possibility of recurrent growth, and the uncertain late effects of post-operative or post-irradiational scarring on the opposite ureter, there is, in these women, an incentive to make a special effort to preserve salvageable renal tissue, particularly since the poorer kidney of to-day may be the better—indeed the only—kidney of to-morrow (Figs. XIV and XV).

FIG. X : E.B. Stage 2 carcinoma cervix. Eight weeks after radical hysterectomy which was followed by development of uretero-vaginal fistula.

FIG. XI : E.B. Three weeks after right uretero-sigmoid anastomosis. Co-existence of left uretero-vaginal fistula had not been diagnosed. Patient continued to discharge urine per vaginam. Bilateral hydronephrosis.

FIG. XII : E.B. Five weeks after right uretero-sigmoid anastomosis. Improvement on right side. Decreased concentration on left side. Well marked concentration of dye in bowel.

FIG. XIII : E.B. Five months after right and three and one-half months after left uretero-sigmoid anastomosis. Marked improvement with good function both sides. Patient very well fourteen months after last operation.

There can be no doubt that the occurrence of this complication could be reduced by the employment of less radical surgical procedures in our attack on uterine cancer, and by excluding advanced or recurrent cases. This may ultimately prove to be the wiser course, but for the present we are unwilling to compromise in this direction.

There are, however, several points relative to the operation which are worth discussing. All are agreed that gentle handling of the ureter and preservation of the peri-ureteric sheath are essential. The ability to satisfy these *desiderata* increases with increasing experience of the operation.

In regard to the preservation of particular vessels during the course of dissection, there are two difficulties. First, the vascular pattern in the pelvis varies from case to case, and secondly, the pathological state is always liable to make the sparing of this or that branch difficult or impossible.

FIG. XIV : E.G. Recurrent carcinoma of cervix. Pre-operative I.V.P. Shows left hydronephrosis and left ureteric calculus. Right side normal.

We have not been able to clear the nodes around the common and internal iliac vessels and at the same time to preserve the branch from the internal iliac artery to the ureter, while deliberate preservation of the branch from the uterine artery seems to us to interfere with radical extirpation of the paracervical and para-vesical tissues. We do attempt to preserve the branch from the superior vesical artery. As it is common to find the uterine and superior vesical vessels arising from a common trunk we are careful to ligate the former only.

Apart from direct vascular deprivation it seems probable that other factors such as infection, haematoma formation and kinks or pressure points may play a part.

FIG. XV : E.G. Four months after anterior exenteration with uretero-sigmoid anastomosis. Shows right-sided hydronephrosis. On the left side the hydronephrosis has improved since the operation. It may be that ultimately the left kidney will be the better one.

We have noted that reperitonealization of the pelvic floor is apt to leave a tight diaphragm which, it seems likely, might exercise pressure on the ureters. The use of interrupted sutures in place of the continuous variety, which we formerly used, certainly has a significant effect on reducing tension from that cause.

It is obviously impossible to assess the actual effect of these various factors. For what it is worth it can be reported that in our last 37 cases, only two uretero-vaginal fistulae have occurred—both of them in cases of late recurrent growth after failed radiotherapy.

The exact diagnosis of uretero-vaginal fistula in regard to site or sites is not always simple. Preliminary intravenous pyelography after the exclusion of vesico-vaginal fistula may demonstrate a dye-filled cavity. But since this is often more or less centrally placed, it may be difficult on the one hand to be sure with which ureter it communicates, and on the other to exclude bilateral fistulae, though the presence of dilatation of the ureter and/or the renal pelvis may give a strong clue.

FIG. XVI : B.F. Nine weeks after operation. Despite presence of uretero-vaginal fistula, ureteric catheters were easily passed on both sides.

Passage of ureteric catheters with ureterography is the next step. Again there are pit-falls. It is sometimes impossible to catheterize the ureters in the post-operative bladder, since oedema or distortion may completely obscure one or both orifices. The catheter may be blocked by oedema or kinking of the ureter, whilst occasionally it may run freely past the fistula (Fig. XVI). Further aid may be obtained by intravenous dye injection with cystoscopy, and at times by injection of dye into the vaginal orifice of the fistula.

Thus, from the practical point of view, it may at times be difficult to exclude with certainty the existence of bilateral uretero-vaginal fistulae; the importance of this in treatment is self-evident (Fig. XVII).

FIG. XVII : E.B. Three months after radical hysterectomy. Right ureterogram showing dilated right ureter and irregular dye-filled fistulous cavity in pelvis. Presence of a concomitant left uretero-vaginal fistula was not detected.

In regard to treatment our views on the expectant attitude have already been presented.

Aldridge and Mason (1950) state that the results of dilating the ureter in the treatment of hydronephrosis suggest that the trauma of dilatation may actually increase oedema and stenosis and hasten the development of "auto-nephrectomy." We have carried out this procedure in one case and the fistula closed fairly rapidly, but the result is poor, there being little function in the kidney on the affected side, with moderately severe contra-lateral hydronephrosis.

Nephrectomy appears fairly frequently in the literature regarding management of a uretero-vaginal fistula, usually as a final solution where expectancy has failed, and under certain circumstances this is no doubt the most satisfactory treatment. In cancer cases, however, the fate of the other kidney is always uncertain and this step requires far more consideration than it would in non-malignant conditions.

We feel, therefore, that there are good reasons for an early attempt to preserve renal function. If this is to be done it is imperative that operation be carried out before the kidneys are damaged. It is clear that many of these operations have failed primarily because they have been performed far too late to have a reasonable chance of success.

Implantation of a ureter into the bladder would obviously be the desirable choice of operation, but in these cases it is seldom practicable to get enough good ureter above the fistula to enable this to be effected without tension, and consequently in most cases the site of ureteric implantation will be limited to some portion of the bowel. Fortunately this does not seem to cause any serious inconvenience in the female. As far as the fate of the kidney is concerned, that seems to depend to a large extent on its condition at the time operation is undertaken.

SUMMARY

The following tentative conclusions may be drawn:

1. That radical surgical attack on uterine cancer necessarily exposes the ureters to the immediate risk of avascular necrosis and the later risk of involvement in scar tissue.
2. That consequently it is important to perform routine post-operative pyelography.
3. That an attack of pyelonephritis should suggest the possibility of ureteric obstruction and demands prompt and intensive treatment and critical observation thereafter.
4. That the development of uretero-vaginal fistula is almost certain to be followed by ureteric stricture, and thus is to be regarded as the first of a series of inevitable events which will end in damage to or destruction of the kidney unless active steps be taken to avoid these consequences. We consider that the *laissez-faire* policy is inherently dangerous.
5. That in the treatment of these complications in the cancer patient, nephrectomy should, if possible, be avoided.
6. That implantation of the ureter into the bladder is seldom practicable in these cases, and that uretero-colic anastomosis, provided that it is undertaken before irreversible changes have occurred in the kidney, may ultimately prove to be the most satisfactory method of managing these problems when they occur after radical pelvic surgery.

ACKNOWLEDGEMENTS

The views presented in this preliminary report have been developed in co-operation with Dr. Arthur Hill, Honorary Surgeon in Charge of the Radio-Surgical Clinic of the Women's Hospital, to whom I am deeply indebted for his generous help and encouragement.

I wish to express my thanks to Dr. Colin Macdonald, Radiologist to the Women's Hospital, for his painstaking radiographic studies and willingness to make his time and experience available whenever a problem required solution, and to Mr. N. Rosenthal, of the Visual Aids Department of the University of Melbourne, for the excellent reproductions of the X-ray films.

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OVARIAN PREGNANCY

A REPORT OF FOUR CASES

By Ross A. Hayes

Department of Pathology, Alfred Hospital, Melbourne

OVARIAN pregnancy is a rare condition, occurring in less than 1 per cent. of all ectopic pregnancies.

The literature on the subject has been well reviewed by Nattrass (1951), Toplack (1950), and Bernstein and Bernstein (1948). It is intended here merely to place on record four further cases which fulfil the criteria formulated by Spiegelberg in 1878, which are:—

(i) The gestation sac must occupy the normal site of the ovary.

(ii) The gestation sac must be connected to the uterus by the utero-ovarian ligament.

(iii) The fallopian tube on the affected side must be normal and separate from the gestation sac.

(iv) Definite ovarian tissue must be present in several positions within the wall of the sac.

Case Histories

Case 1

M.E., aged 35. Four children—eldest aged 14 years, youngest 6 years. Nothing significant was to be noted in the past history. Menses were regular, a twenty-eight day cycle. She was first seen on 20th November, 1949, when her period was one week overdue. She was apparently pregnant, aborted, and was curetted on 27th December. She was next seen 25th November, 1951, after she had collapsed at home with severe abdominal pain whilst at stool. On examination she was found to have some tenderness in the right iliac fossa and in the right fornix. Last normal menstrual period 30th October. At this stage there were no signs of intra-abdominal haemorrhage and in the absence of any definite diagnosis she was admitted to hospital and observed. The tenderness gradually subsided and on 11th December the abdomen was opened through a lower right paramedian incision. There was some blood-clot in the pelvis, the uterus was enlarged and the right ovary was adherent between the posterior abdominal wall and the posterior wall of the uterus, and was noted to be enlarged, cystic, and haemorrhagic. The uterus

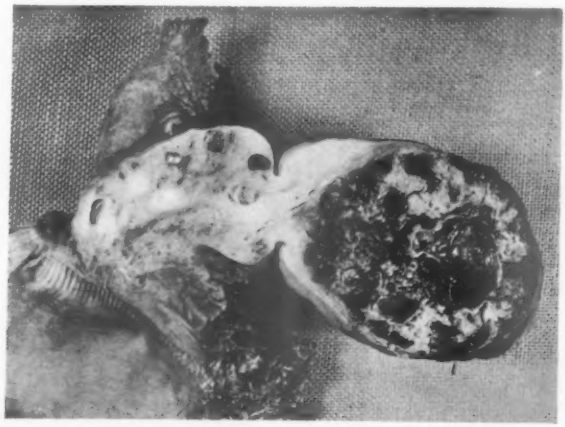

FIG. I. Photograph of uterus, tube and ovary of Case 1. The ovary is thinned out around the pregnancy and is notched by a surgical ligature.

and both ovaries were removed. One pint of blood was given to speed convalescence which was uneventful.

Pathological Report

Macroscopic examination. The uterus was slightly enlarged. The Fallopian tube on the affected side was normal. The ovary, 3 x 7 x 4 cm., had been artificially notched by a surgical tie (Fig. I). The normal ovarian tissue was thinned out around a fleshy haemorrhagic mass of tissue containing a central cavity, 1 cm. in diameter, lined by a glistening membrane. No foetus could be identified.

Case 2

A.C., aged 31 years. One child, aged 10 years. For the last three to four years she had had amenorrhoea spasmodically for three to four months at a time. Five months prior to operation she had an attack of severe cramping lower abdominal pain, worse on left side, which lasted two weeks. No vaginal discharge. She had had some dysmenorrhoea since. On examination of abdomen, some tenderness and sense of resistance was felt in the left iliac fossa. Cervix was not enlarged and was freely movable. A mass was felt in the left fornix. The abdomen was opened through a lower midline incision and a large cystic left ovary was seen and removed. The remaining pelvic organs appeared normal.

FIG. II. Low power photomicrograph of a section of portion of the specimen shown in Fig I, showing normal ovarian stroma with atretic follicles and areas of haemorrhage and many chorionic villi. (x 3)

Microscopic examination. The uterus showed well-developed decidual reaction. The left ovary contained a *corpus luteum*. The right ovary showed typical trophoblast and chorionic villi and, at several places around the edge, normal ovarian stroma with follicular cysts and atretic follicles (Figs. II and III).

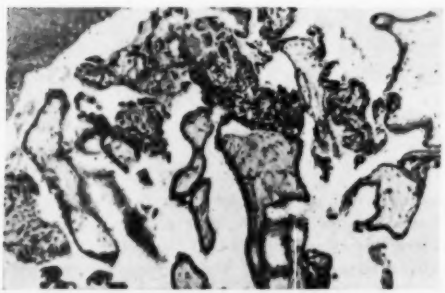

FIG. III. High power photomicrograph of the chorionic villi of Case 1. (x 80)

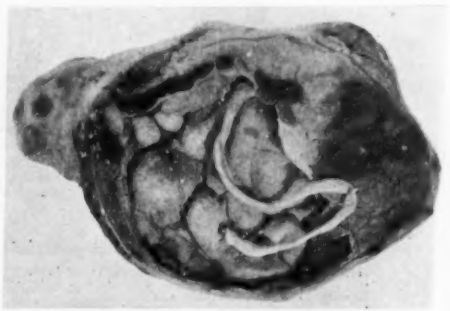

FIG. IV. Photograph of the ovary from Case 2 showing the ovary thinned out around the haemorrhagic area with the central cavity and attached cord.

Pathological Report

Macroscopic examination. The ovary measured 10 x 6 x 7 cm., and consisted of a lobulated mass of haemorrhagic tissue and a central cavity, 5 cm. in diameter, containing a macerated foetus attached by a cord 10 cm. long (Fig. IV).

Microscopic examination. Normal ovarian tissue containing follicular cysts and atretic follicles surrounded a haemorrhagic zone containing necrotic "ghost" chorionic villi.

Case 3

A nulliparous woman, aged 24, who had been married for eight months. She had a miscarriage at two and a half months gestation for which she was curetted. The endometrium was not examined microscopically. She did not menstruate following this operation and six weeks later, on examination, was found to have a mass thought to be a left ovarian cyst. The following week she was operated on and the enlarged left ovary was removed. The remaining pelvic organs were inspected and appeared normal. She later divorced her husband and re-married but had no children some five years later.

FIG. V. Photograph of the ovary from Case 3 showing the central cavity with the foetus *in situ*.

Pathological Report

The specimen consisted of an ovary, 5 cm. in diameter. The major portion was dark in colour with large vessels running on the surface and some nodules up to 2 mm. in diameter protruded above the generally smooth surface.

Macroscopic section. There was a cavity, 3 cm. in diameter, lined by a smooth membrane. It contained a foetus, 0.8 cm. in length. The wall of the cavity was 0.5-1 cm. in thickness with large sinuses present. A small portion of ovarian tissue was seen at one pole (Fig. V).

Microscopic examination. To one side of the section was ovarian stroma in which several atretic follicles were seen. As the cavity was approached, there were numerous large, thick-walled vessels which had marked hyaline change in their intimal portions. The cavity was lined by blood clot in which many degenerate chorionic villi were seen. On the surface was a well-defined amniotic membrane.

Case 4

A.H., aged 30 years, had 4 normal living children; the youngest aged 10 years. She had had 2 miscarriages since the last child. She presented on 27th October, 1941, complaining of nagging pain in the left iliac fossa for eighteen months and some frontal headaches. Her periods had been regular until six months prior to admission when she had developed pain across the lower abdomen which had lasted two days and was associated with some vomiting. Since then she had had a black vaginal discharge, somewhat offensive, with some clots. On examination, the uterus was thought to be slightly enlarged. A laparotomy was performed. The uterus, right and left tubes appeared normal. The left ovary was seen to be about the size of a golf ball and was removed with the left tube. The abdomen was closed and convalescence was uneventful.

Pathological Report

Macroscopic examination. The ovary was 3.5 cm. in diameter and contained a mass of blood-clot. There was a small cavity in the centre of this clot. There were no distinguishing features of the wall of the cavity. The tube appeared normal.

Microscopic examination. Section through portion of the ovarian mass revealed normal ovarian stroma with an atretic follicle alongside which there was a mass of blood-clot; the edge of which was becoming organized. In the blood-clot were the remains of numerous degenerate chorionic villi. Foetal parts were not seen.

DISCUSSION

Primary ovarian pregnancy is one of the rarest types of ectopic pregnancy. In 920 cases of ectopic pregnancies at the Women's Hospital, Melbourne, over a ten-year period, there have been 2 cases of primary ovarian pregnancy (Forster, 1952; Nattrass, 1951). These cases are indistinguishable, clinically, from the more common tubal pregnancy.

One interesting feature in the clinical histories of three of these cases is the period of sterility preceding the ovarian pregnancy. This has been emphasized by Bernstein and Bernstein (1948) and others. The aetiology

of the condition is still a matter for conjecture and has been discussed fully by Nattrass, but from these cases there is no evidence in favour of ovarian endometriosis.

SUMMARY

Four cases of primary ovarian pregnancy have been described which fulfil the rigid criteria for such a diagnosis. The history of previous sterility is emphasized.

ACKNOWLEDGEMENTS

I wish to thank Mr. R. Fowler, Dr. W. Phillips and Dr. Daly Smith for their permission to publish these cases and for the clinical details which they have supplied. I would also like to thank Dr. J. Funder, Dr. J. D. Hicks and Dr. A. V. Jackson for the pathological material.

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THYROGLOSSAL CYST

By Rowan Webb

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THIS report records the results of a review of 41 patients who were operated on for thyroglossal cyst or fistula at the Royal Melbourne Hospital during the period 1920-1950.

General Features

Of these 41 patients, 21 were males and 20 females. Their ages ranged from 2 to 66 years, the average age at operation being 30 years, and over half of the cases presented between the ages of 20 and 50. In only 4 cases did symptoms date from birth. Sixteen patients had noticed symptoms for less than one year, and three-quarters of the cases gave a history of less than ten years. A common mode of presentation was with sepsis in a midline swelling in the neck, treated by incision, which was followed by a persistent sinus or by recurrent swellings.

Larger series have been recorded in the literature, notably by Marshall and Becker (1942), who reported 348 patients treated at the Lahey Clinic, and Gross (1940), also from Boston, who had studied 198 cases. Both these papers dealt with the clinical aspects of the condition rather than with details of surgical pathology. Indeed, most of the standard textbooks of pathology devote little space to the histology of the condition, and give little indication of the variety of tissues which may be found. The present study revealed not only that several varieties of lining epithelium may be seen—often in the one section — but also that a number of different tissues can occur in the walls of the cysts and the ducts.

Surgical Pathology

In a careful study of three cases published in London in 1894, Durham observed that the thyroglossal duct may be branched, may possess diverticula and, in parts, may consist of solid epithelial cords. He found a lining of "respiratory" epithelium in two of his cases, and suggested that its presence was of considerable diagnostic value. On two occasions he observed tissue resembling thyroid vesicles in the wall of the duct.

FIG. I. Photograph of a bilocular thyroglossal cyst.

It was not possible to determine the frequency of occurrence of branching of the ducts or of diverticula in the present series, since in many cases, single sections only were available, rather than serial sections or dissections of macroscopic specimens. Multiple lumina were occasionally seen, and while many were obviously sections through one tortuous duct, others showed variations in calibre and epithelium in keeping with the presence of branching or duplication of the duct. Thyroglossal cysts are usually unilocular but may be bilocular (Fig. I) or even multilocular.

The Epithelium Lining the Cysts

Ten specimens were so altered by previous suppuration that no epithelium could be detected. The remaining 31 cases were remarkable for the range of tissues in both the lining and the walls of the ducts.

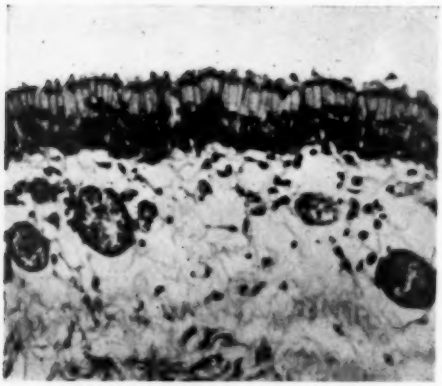

FIG. II. Photomicrograph of portion of the wall of a duct showing pseudo-stratified ciliated columnar epithelium. (x 225)

The commonest lining epithelium was a mixture of simple cuboidal and columnar epithelium (17 cases). There was commonly one layer, but in some places the epithelium was stratified, as many as eight or ten layers being present. The height of the epithelium varied from place to place in each section, as also the number of layers of cells.

TABLE 1

Distribution of types of epithelium lining thyroglossal cysts and ducts. In 10 of these cases the epithelium in considerable parts of the wall had been destroyed leaving only a connective tissue surface.

Type of Epithelium	Number of Cases
Cuboidal and columnar	17
Pseudo-stratified ciliated columnar	15
Stratified squamous	10
Total	42

Well-differentiated pseudo-stratified ciliated columnar epithelium was found in almost half (15) of the sections which contained epithelium (Fig. II). Well-developed goblet cells were also conspicuous in these cases.

TABLE 2

Distribution of types of tissue in the wall of thyroglossal cysts and ducts.

Type of Tissue	Number of Cases
Thyroid vesicles	15
Lymphoid tissue	5
Salivary glands	2
Sebaceous glands	2
Total	24

The third common type of lining was stratified squamous epithelium (9 cases), with well-defined prickle cells, and a variable degree of cornification of the surface cells (Fig. III). This last feature, in most cases, was not gross.

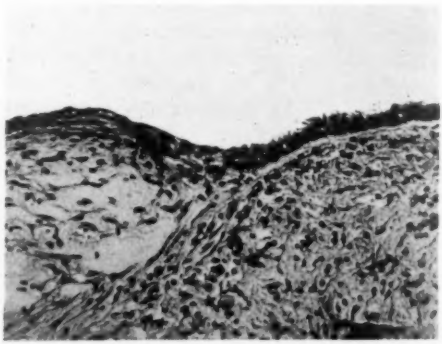

FIG. III. Photomicrograph of the wall of a thyroglossal duct showing both stratified and "respiratory" epithelium. (x 150)

Less commonly, parts of the wall were lined by a single layer of flattened epithelium, and on a few occasions areas were seen which appeared to be lined simply by granulation tissue—doubtless the result of infection.

Epithelium in the Wall

Thyroid tissue was found frequently in the wall of the duct or cyst (15 cases). Well-defined vesicles, usually small, were readily recognized and were arranged either in clumps or were more sparsely scattered (Fig. IV). In most cases the epithelium lining these was cuboidal and colloid was present.

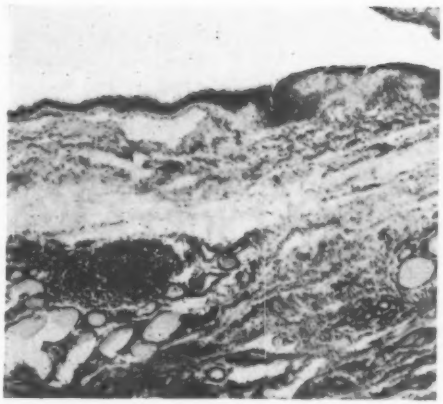

FIG. IV. Photomicrograph of the lining and wall of a thyroglossal duct showing thyroid vesicles and lymphoid tissue. (x 50)

Two specimens contained clumps of salivary glandular tissue in their walls (Fig. V), this consisting of both serous and mucus-secreting cells, and salivary ducts.

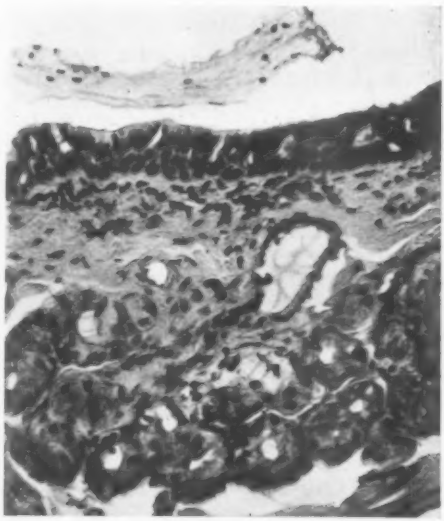

FIG. V. Photomicrograph of section of the wall of a thyroglossal cyst showing salivary gland tissue. (x 225)

Small mucous glands were frequently seen, particularly in association with "respiratory" epithelium and goblet cells were numerous. Well-developed sebaceous glands were also found in relation to stratified squamous epithelium (Fig. VI).

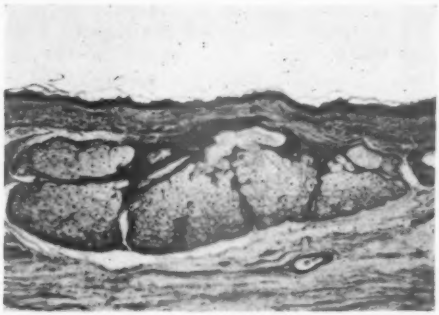

FIG. VI. Photomicrograph of a section of the wall of a thyroglossal cyst showing stratified squamous epithelium and sebaceous glands. (x 50)

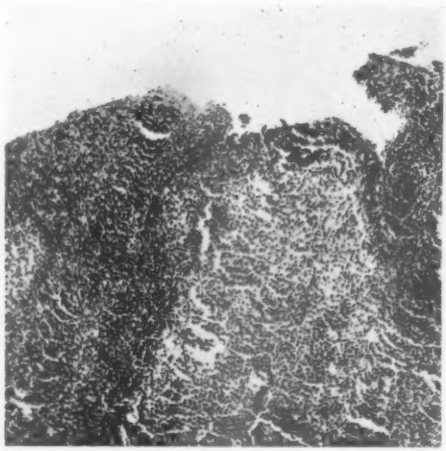

FIG. VII. Photomicrograph of a section of the wall of a thyroglossal duct showing lymphoid tissue in wall. The poor demarcation of squamous epithelium from lymphoid tissue is demonstrated. (x 75)

Clumps of lymphoid tissue were present in the sub-epithelial tissues in a number of sections, even in the absence of inflammation. This was noteworthy in 5 cases, germinal centres being prominent. The lymphoid tissue is closely related to the epithelium and, in many cases, there is a very poor demarcation between the two (Fig. VII). This observation is yet another example of a

common histological finding in both normal and pathological conditions in the neck and mediastinum. The phenomenon is particularly well seen in such conditions as branchial cyst, adenolymphoma of the parotid gland, and in some diseases of the thyroid gland and the thymus. This subject has recently been discussed in detail by Heinz (1950).

DISCUSSION

The thyroid gland develops from a median diverticulum, which appears immediately caudal to the *tuberculum impar*, corresponding in site to the *foramen caecum* of the fully developed tongue. Persistence of part of this downgrowth might therefore be expected to reproduce thyroid tissue, and indeed this is frequently the case. Ectopic tissue may be found anywhere along the track, either alone or in association with thyroglossal cysts or persistent duct. It is important to realize, however, that instead of thyroid tissue, other types of epithelium may be found. The macroscopic appearance of the cyst will vary accordingly, and the contents in an uninfected case may be clear thin fluid, thin or thick mucus, or sebaceous material.

Detection of cuboidal or columnar cells resembling thyroid epithelium is by no means essential for the diagnosis of thyroglossal remnants, which is made on topographical grounds, and these structures can still be confidently identified even if the sections from a clinically typical case reveal, not thyroid epithelium, but "respiratory" or stratified squamous epithelium, or merely a single layer of flattened cells.

The problem of multipotency of tissues is commonly encountered in various parts of the body and it is important that it should be recognized here as a matter of practical importance. Failure to appreciate its influence on adult tissues may lead to unnecessary and artificial difficulties in diagnosis.

The common occurrence of epithelial elements outside the lumen of a tract and the possibility of ramification of the ducts emphasize the need for the removal of an adequate margin of tissue around the duct if the excision is to be complete. The varying relations of the duct to the body of the hyoid bone, and the necessity for the removal of this structure, when the tract is excised are well known.

SUMMARY

A study of 41 cases of thyroglossal cysts and ducts is presented.

These cases were distributed almost equally between the sexes, and usually first gave rise to symptoms in early adult life.

The different types of epithelium which may be found lining, and in the walls of, these structures are described. These range from simple epithelia, through "respiratory," thyroid and salivary tissue to squamous epithelium.

The clinical significance of these findings is discussed.

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A CASE OF CHORDOMA WITH PULMONARY METASTASES

By R. B. MAYNARD

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CHORDOMA, a tumour arising in the spine and histologically composed of characteristic cells which resemble those of the pulp of the intervertebral disc, is an uncommon growth. It is invasive in its development but only occasionally gives rise to metastases.

In view of this rarity a case of chordoma of a lumbar vertebra with a pulmonary metastasis is recorded here.

CASE HISTORY

A male patient, aged 46, was admitted to hospital on 2nd February, 1951, with a history of gradual onset of lower lumbar backache. It had been present for about eleven weeks and for three weeks prior to admission pain had radiated down the right thigh to the inside of the knee. Physical examination failed to demonstrate any physical abnormality other than tenderness in the lower lumbar region of the spine and inability to sit up because of pain at the site of tenderness. Reflexes and sensation were within normal limits.

X-ray examination showed:—

(a) a destructive process in the body of the fourth lumbar vertebra with virtual disappearance of the body (Fig. I). The upper dorsal spine appeared to be osteoporotic but there was no definite lesion in this region;

(b) a solitary area of rarefaction in the left parietal bone posteriorly and a small punched-out area in the shaft of the right radius close to the interosseous membrane;

(c) normal intravenous pyelogram.

It was concluded that the possibility of myelomatosis could be discarded but that renal or thyroid malignancy should be considered in the diagnosis.

Ancillary investigations

Blood examination: haemoglobin 11 gm. (100 ml.); red blood cells 3.5 million per cubic mm.; white blood cells 8,000 per cubic mm.; erythrocyte sedimentation rate 132 mm. in 1 hour (Westergren).

Urine examination: no albumen or proteose was found but an occasional red blood cell and hyaline cast was seen in each high power field (centrifuged specimen).

Biochemical examination: blood urea 35 mgm.; urea clearance 100 per cent.; serum protein 6.65 gm.; serum globulin 2.65 gm.; acid phosphatase 2.1 units; alkaline phosphatase 25 units; inorganic phosphate 3 mgm.; serum calcium 11.4 mgm.

Wassermann test gave a negative reaction.

FIG. I. X-ray photograph of the lumbar spine showing replacement of the fourth lumbar vertebra by non-opaque tissue. (Lateral view)

The patient was nursed in a plaster bed and a course of deep X-ray therapy was given but he died on 30th March, 1951.

Post-mortem examination

The relevant findings were that "in front of the third and fourth lumbar vertebrae there was a

lobulated projecting tumour consisting of fairly large lateral lobes with an isthmus joining them in front. This isthmus measured 6 cm. from above down and the tumour 11 cm. transversely. The right lateral lobe measured 7.5 cm. from above down and the left approximately 5 cm.

On section, the projecting lateral nodule on the right showed a pale, mucoid, neoplastic tissue. There was no haemorrhage and little evidence of necrosis. The mobility of the second lumbar vertebra on the lower part of the specimen was in keeping with the gross destruction of the fourth lumbar vertebra. The sacrum, third, fourth and fifth lumbar vertebrae and portion of the ileum on each side were removed *en bloc* for further examination.

There was a small, white hemispherical deposit on the pleural surface of the left lung, on the upper portion of the upper lobe."

Remainder of autopsy findings were not relevant to this report. The site of the radiological lesions of the skull and right radius showed bone morphologically similar to that of neighbourhood and there was no suggestion of tumour.

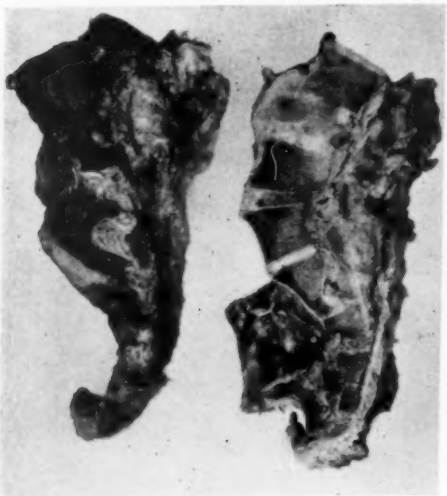

FIG. II. Photograph of the specimen of a chordoma of the fourth lumbar vertebra, showing replacement of the vertebra by tumour tissue.

Macroscopic examination

The fourth lumbar vertebra was almost replaced by yellowish tumour mass continuous anteriorly with the abdominal tumour mass (Fig. II).

Both cranially and caudally there was invasion of the intervertebral discs and the body of L3 by mucinoid tumour substance which replaced normal bony trabeculae. Extension dorsally had considerably narrowed the lumen of the vertebral canal in the region of L3, 4 and 5, the coverings of the cord in this area being intact and uninvaded. Fairly extensive infiltration was demonstrable in the ligaments and muscles adjacent to the affected area.

The intervertebral disc between L5 and S1 was largely replaced by a pale reddish-brown tissue which appeared to be homogeneous but of invasive habit involving the bodies of S1, 2 and 3 and extending dorsally to involve laminae, spinous processes, and loose tissues of the sacral region.

Histological examination

The initial examination was by means of small sections representative of the whole lesion but later a complete slice was decalcified and examined.

The main bulk of the tumour consisted of a fairly loose network of physaliphorous cells amongst which were some areas of denser, almost solid appearance.

There was some evidence of invasion of the fibrous capsule which in the main was well delineated and preserved. Studied in more detail the loosely packed areas showed considerable variation in structure, size of cell, and also in the amount of mucinoid material both free and in the cytoplasm, this latter characteristic often giving the cells a vacuolated appearance. Areas of extracellular mucin could be demonstrated even in routine haematoxylin and eosin-stained specimens.

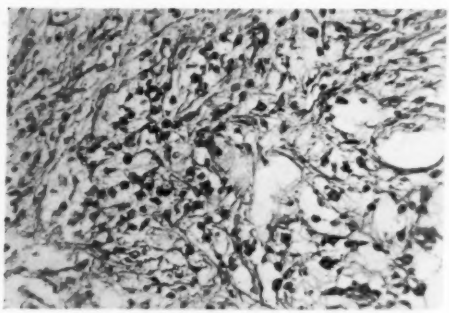

FIG. III. Photomicrograph showing the cellular character of the tumour. The cells show the mucin accumulations both within and without the cells. The cells are large. (x 200)

Selective staining amply confirms the presence of both intra- and extra-cellular mucin (Figs. III and IV), although in our hands the muci-carmine stain was not as effective as the standard stains of haematoxylin and eosin and Masson in demonstrating the mucinous intra-cellular components of the tumour cells.

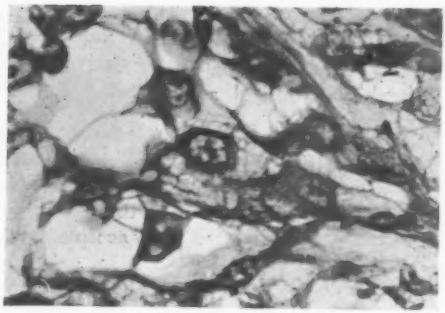

FIG. IV. Photomicrograph of a portion of the primary tumour. The large size of the cells is shown and there are droplets of mucin in the protoplasm and outside the cells. (x 1500)

The extra-cellular mucin in places is the predominant feature with islets of cells scattered throughout but in general this growth is rather more cellular than is usually described and in the more cellular areas the individual elements are polyhedral and arranged in cords and clumps. This appearance is totally unlike that of the loosely packed areas where the cells vary from small polyhedral to large, sprawling cells with every conceivable diversity of shape and size. Many multinucleated cells occur and in cellular areas there is also evidence of mitosis although this is not a prominent feature.

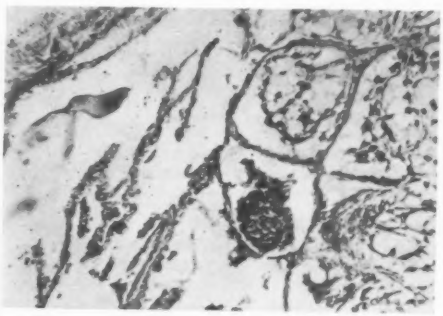

FIG. V. Photomicrograph of a section from a nodule in the lung. (x 200)

Histologically the pulmonary metastasis shows a somewhat more even cell distribution than the parent tumour but none-the-less has the typical chordomatous appearance and well developed "physaliphorous" cell characteristic (Figs. V and VI). The nodule is small and there is a fine surrounding fibrous condensation or pseudo-capsule.

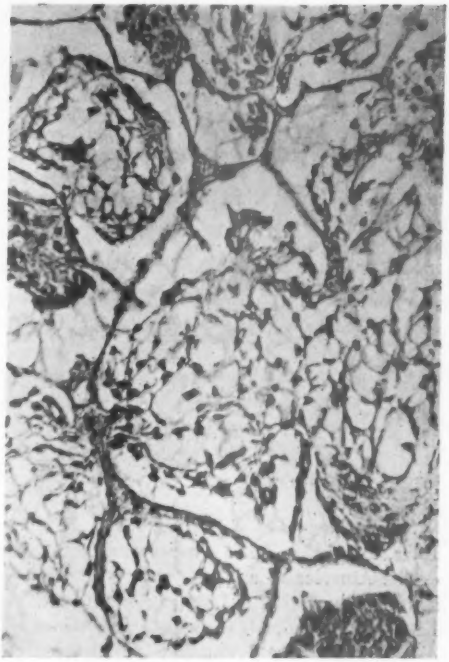

FIG. VI. Photomicrograph of a more cellular portion of one of the lung metastases. (x 225)

DISCUSSION

Tumours of chordomatous type had been reported since 1856 but so rarely that M. J. Stewart (1922), in his first review, was unable to collect more than 26 acceptable examples of the condition up to that date. Following the initial review by Stewart, reports became somewhat more frequent so that in 1926, in collaboration with Morin, he was able to collect a further 31 cases.

Isolated reports continued to appear and in 1941 Harvey and Dawson collected 240 acceptable examples of the condition and it was possible to analyze the figures from the usual statistical standpoints. In the same year a review by Horwitz (1941) of 245 cases appeared.

The tumours are situated in three main sites; the spheno-occipital (cranial) region, the vertebral column and the sacro-coccygeal region.

Harvey and Dawson (1941), in the 240 cases, showed a distribution:—

Cranial	37 per cent.	(88 cases)
Vertebral	12 per cent.	(30 cases)
Sacro-coccygeal	51 per cent.	(122 cases)

Horwitz (1941), in 245 cases, showed a similar distribution:—

Cranial	33 per cent.	(81 cases)
Vertebral	12 per cent.	(29 cases)
Sacro-coccygeal	55 per cent.	(135 cases)

As the reviews of Stewart and Morin (1926), Harvey and Dawson (1941) and Horwitz (1941) were so complete, further analysis of the cases from 1856 to 1942 would be redundant.

Search of the literature from 1941 to 1951, using the *Index Medicus* and the index of the American Cancer Society as the initial sources, show that a further 68 reports have been published. These describe 41 cases distributed in the proportion of 15 vertebral and 26 sacro-coccygeal which deviates considerably from the proportions shown above.

The presented case must be placed in the vertebral group even though the bulk of the tumour had disseminated by local spread through sacrum and adjacent tissues.

In all reports there is evidence of great invasiveness and local malignancy but very few articles illustrate metastases or even lymphatic spread. Indeed, the rarity of metastatic deposits is so well accepted that in the report of Penkert (1941) a pelvic tumour is considered to be possibly chordoma because of the lack of metastases. Only a few of the recently reported cases have described metastases though probably this is due to the predominantly large contribution from surgical and radiological journals in which case there is seldom any autopsy evidence.

Willis (1948) describes metastases in seven cases, the dissemination being to lymph glands, liver, lungs and skin. Of the purely sacro-coccygeal chordoma Gentil and Coley (1948) collected 14 reports in which metastases are described and this in a total of 128. The sites of deposit are lymph nodes, lung, liver, kidney, adrenal, muscle, heart, skin and thyroid.

TABLE 1

Author	Site	Description of Case
von Güthert, H. and Henkel, H. (1941)	Metastasizing chordoma of lumbar vertebrae.	Woman, aged 50. Multiple metastases in lung.
Faust, D. A., Gilmore, Jr., H. R. and Mudgett, C. S. (1944)	Sacro-coccygeal tumour.	Male, aged 23. 12 months history. Origin from sacrum. Metastases in lung, muscle, left adrenal. Post-mortem diagnosis.
Robins, S. L. (1945)	Lumbar vertebral chordoma.	Male, aged 58. Unusually large sacro-coccygeal tumour. Metastases to regional and inguinal lymph glands, liver, lungs, pleura and skin of right leg and thigh. Histological illustrations typical.
Morris, A. A. and Rabinovitch, R. (1947)	Malignant chordoma of lumbar region. Metastases to brain, lungs, pancreas, sacral axillary and iliac lymph glands.	Female, aged 27. Original diagnosis based on biopsy 26 months before visceral pleural nodules, metastases right and left lungs. Nodule in peri-pancreatic soft tissues, axillary and iliac lymph nodes. Extensive destruction of bone of left side of skull with large dural nodules and large irregular mass 6 x 4 cm. attached firmly to underlying brain.

Table showing cases of chordoma with metastases from 1941-1951.

The incidence of metastasizing tumours in the 1941-1951 series was only four (Table 1) in a total of 41 cases and this conforms with the figures of Gentil and Coley (1948) who collected 14 in 128 (approximately 9 per cent.); but it is doubtful whether this represents the true incidence of metastases for this cannot be correctly gleaned from reports which are mainly clinical. Chordomas are, in general, of such long history that years may pass from the initial symptom to death (6.4 years on an average with maximum of 19 years—[Stewart and Morin, 1926]). Further, it is doubtful if all case histories are closed by published autopsy with consequent full record of metastases.

Summary

A case of chordoma with pulmonary metastases is presented and accompanied by some observations on the incidence and site of true metastatic deposits.

Acknowledgements

The post-mortem examination was carried out and the diagnosis made by Professor M. J. Stewart during a visit to Melbourne. This case is presented with the consent of the Chairman of the Repatriation Commission. The assistance of Miss H. Wischusen with the photographic reproduction and Miss Cuzens in searching records is gratefully acknowledged.

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SPREADING BACTERIAL GANGRENE OF SKIN

WITH REPORT OF ONE CASE

By J. M. Yeates

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A REVIEW of the literature shows that two types of infective gangrene of the skin are recognized:—

(1) A chronic form, well known under the name of symbiotic or synergistic gangrene.

(2) An acute form, not well known, and referred to as acute haemolytic streptococcus gangrene (Meleney).

These two varieties of gangrene have this in common—that they are caused by infection of the subcutaneous fat by micrococci.

Both these infections can be controlled by the use of certain antibiotics.

Historical

(a) Chronic Gangrene

In May, 1924, Cullen described a case in which a progressively enlarging ulcer followed drainage of an appendical abscess. On 7th November, 1922, he cut away the indurated and reddened area with a cautery. Fifteen days later he noted that "the infection seems to be clearing."

In 1924, Christopher reported a case following drainage of an empyema. Pain was most pronounced and at times the patient could not bear to be touched. Cure followed excision of the ulcer margins (10 operations).

In 1926, Brewer and Meleney each reported a case, both following appendical abscess. At the same time they demonstrated the responsible organisms (non-haemolytic micro-aerophilic streptococcus and haemolytic staphylococcus) and were able to produce characteristic ulcers in experimental animals by simultaneous injection of these organisms.

In 1930, Poate published a case report. This appears to be the first recorded case in a British Journal. In 1933, Scotson was responsible for the first published account in England.

Meleney (1945) and his co-workers reported two cases cured by penicillin without operation. It is of particular interest that a third case required excision because the presence of *Escherichia coli* produced penicillinase which inactivated penicillin.

In 1948, Meleney showed that bacitracin was an effective therapeutic agent. In two cases the infection was controlled in forty-eight hours.

(b) Acute Gangrene

This condition appears to have been first described in 1924 by Meleney who also isolated the causative organism (haemolytic streptococcus). An excellent account with numerous references is given in Meleney's text-book. Unfortunately none of the references is available in Sydney.

Case Report

On 31st January, 1950, a thin woman, aged 55, was injured in a bus accident.

Examination in hospital some hours later showed that the left leg had borne the brunt of the injury. There were extensive abrasions and bruising from knee to ankle. The foot was in a position of abduction. Near the inner aspect of the ankle joint there was a linear wound 1 inch long, through which blood-stained, fatty tissue protruded. An X-ray showed that both malleoli and the neck of the fibula were fractured.

At 8.00 p.m., operation was performed under general anaesthesia. After the preliminary cleansing of the entire leg the soiled fat and some skin were excised from the small wound near the ankle joint. There was no evidence of gross contamination.

Penicillin powder was applied and the wound was sutured. The deformity was then reduced by manipulation. Dressings and plaster of Paris were then applied. Penicillin injections were commenced from the day of admission.

CLINICAL FEATURES

It will be most convenient to arrange the clinical features of the two types in tabulated form:—

	CHRONIC FORM	ACUTE FORM
ORGANISMS	Micro-aerophilic streptococcus and Haemolytic staphylococcus	Haemolytic streptococcus
ANTECEDENT LESION	Deep abscess of body especially appendix or empyema	Superficial wound of limbs (May be trivial or unrecognized)
DURATION OF SPREADING	Months	Days
PAIN	A prominent feature May be intolerable	Not prominent May be absent
FEVER	Little	Often to 102°F.
ONSET	Insidious	Sudden
PATHOLOGY		Thrombosis of vessels in the fat
ANAEMIA	No	(Marked in this case)
TREATMENT		
(a) Medical	Penicillin — bacitracin	Sulphonamides and penicillin Bacitracin
(b) Surgical	Radical excision	Incision for drainage

For the next few days progress was normal except that the temperature was slightly raised. During the next week the temperature became more elevated with occasional peaks of 102°F. The penicillin dosage was greatly increased, sulphonamides and streptomycin were added.

Meanwhile the patient's general condition was very good. She complained of no pain in the injured leg. The colour, movement and sensation of the toes were normal.

On the 13th February the plaster of Paris was removed. It came as a surprise to find that there was fluctuation under the skin from knee to ankle and that the skin itself was gangrenous or nearly so.

Long axis incisions were made with the release of much blood-stained purulent fluid. The wounds were then irrigated with hydrogen peroxide and large dressings were applied.

The next day *Escherichia coli* and *Bacillus subtilis* were found in the culture taken from the fluid. A second culture grown in both aerobic and anaerobic conditions produced the same organisms.

The temperature dropped soon after operation, but chemotherapy was continued as before.

On 20th February, it was obvious that the skin of the leg was doomed as it was black and insensitive. It was therefore completely excised under general anaesthesia. When this was done it could

FIG. I. Photograph of leg immediately prior to first skin graft.

clearly be seen that the deep fascia and muscles were quite intact, contrasting sharply with the devastation of the superficial tissues. Seven days later the wound was covered with good quality granulation tissue. On this day, therefore, large split skin grafts were cut from the opposite thigh and applied to the raw areas. These were particularly well received, but gaps were filled on 6th March and again on 13th March. From now on progress was steady.

FIG. II. Photograph of leg taken on day of visit to follow-up clinic (1952).

On 19th April, X-ray showed firm union of the fracture in good position (despite lack of proper immobilization). Walking was now commenced. The wounds were all healed except for a few crusty areas.

A follow-up examination on 19th March, 1952, showed that the skin was in good condition and the patient walking with a scarcely perceptible limp.

During the critical stages anaemia was pronounced as will be seen from the following facts.

Date	Haemoglobin	Blood given
13th February	7.7 gm.	1½ litres
21st February	10.8 gm.	1 litre
27th February	?	1 litre
1st March	14.6 gm.	—
7th March	10.7 gm.	—
30th March	11.2 gm.	—

DISCUSSION

It will be seen from the table that the case described resembles very closely the acute form of skin gangrene.

It may be argued that such a diagnosis is untenable in the apparent absence of the haemolytic streptococcus.

There are, however, two possible explanations for this deficiency:—

(a) That such an organism was present and active during the vital early days when the penicillin dosage was relatively small and that later (when the culture was made) it had been destroyed by increasing doses of penicillin and sulphonamides.

(b) That the swabbing for culture purposes was taken from the centre of the abscess rather than from the edge of the lesion where activity is more likely.

The fact that *Escherichia coli* were present is of considerable interest as this organism is known to produce penicillinase, which tends to inactivate penicillin.

This may have been the reason for the failure of routine doses of penicillin to control a possible streptococcal infection.

SUMMARY

1. Two forms of spreading bacterial gangrene of the skin are defined.

2. A brief review of the literature on this subject is given.

3. The two types of gangrene are contrasted in tabular form.
4. A case report is given.
5. In discussing this case, reasons are given for making a diagnosis of acute haemolytic streptococcal gangrene.

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The title-page of the 1559 edition of the *De Re Anatomica*. This is the only illustration in the book.

The College Library

THE *DE RE ANATOMICA* OF REALDUS COLUMBUS

By K. F. Russell

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WHEN Vesalius retired from the Chair of Anatomy in Padua in 1544 his successor was Mattheus Realdus Columbus who had acted as deputy to Vesalius during the latter's period of leave in the previous two years.

Columbus was born in Cremona about 1516 and studied medicine in Venice and Padua; he was a student and later an assistant of Vesalius and in 1544 he occupied the Chair at Padua. He did not remain there long, however, for in 1545 he was appointed the first Professor of Anatomy at Pisa and he remained there until 1548 when he accepted the Chair of Anatomy in Rome which he occupied until his death in 1559.

During his lifetime he was a friend of Michelangelo and it is probable that the painter studied much of his anatomy and did his dissections of the body with the help of Columbus.

The only work that Columbus published was his *De Re Anatomica* and he died while it was being printed, its publication being completed by his two sons. The book owes much to Vesalius; indeed it would seem that Columbus has based his text on that of the *Fabrica* but, in spite of his debt to his master, Columbus does not hesitate to criticize Vesalius and attempts to show that, in fact, the real master is Columbus himself. The only illustration in the book is the woodcut title showing the author carrying out a dissection and this woodcut is obviously based on the famous one in the *Fabrica*.

The text is divided into the following fifteen chapters:—the bones, the cartilages, the ligaments, the skeleton, the muscles, the liver and veins, the heart and arteries, the brain and nerves, the glands, the eyes, the viscera, the formation of the foetus, the skin, on vivisection and finally on those things rarely found in anatomy. The matter in each chapter is briefly but accurately discussed and no opportunity is lost to point out the errors of former writers. In spite of certain shortcomings the author does show evidence of original thought and his descriptions of the mediastinum, pleura, peritoneum and eye are more accurate than former writers. In the last chapter he gives an early reference to gallstones when he describes the post-mortem on St. Ignatius of Loyola at which he was present.

However, this book must always be important in the history of anatomy for, on page 177 in the chapter on the heart and arteries, Columbus prints a clear and concise account of the pulmonary circulation and this is the first time that such an account had been available for general reading. It is true that Michael Servetus (1511-1553) had described the pulmonary circulation in his *Christianismi restitutio* but when he was burned at the stake in 1553 all copies of his book, except three, perished with him.

The description given by Columbus, as translated by Foster (1901) states:—

> Two cavities, that is two ventricles, are present in the heart, not three as Aristotle thought, of these one is on the right side, the other on the left. The right is much larger than the left. The right contains the natural blood, but the left the vital blood. It is very interesting to observe that the substance of the heart surrounding the right ventricle is very thin but on the left side is very thick; and this is so arranged on the one hand in order to keep up the balance and on the other to prevent the vital blood which is exceedingly thin from transuding out of the heart. Between these ventricles there is placed the septum through which almost all authors think there is a way open from the right to the left ventricle; and according to them the blood is in the transit rendered thin by the generation of the vital spirits in order that the passage may take place more easily. But these make a great mistake; for the blood is carried by the artery-

like vein to the lung and being there made thin is brought back thence together with air by the vein-like artery to the left ventricle of the heart. This fact no one has hitherto observed or recorded in writing; yet it may be most readily observed by anyone.

It is to be noted that Columbus claims the pulmonary circulation as his own invention and it is said that he copied it from Servetus without acknowledgement, and this may be true but in spite of what is known of Columbus and his tendency to egotism, he was the one who first made available to the world a printed description of the minor circulation and also demonstrated it experimentally and therefore deserves credit.

Actually modern investigation has shown that neither Servetus nor Columbus were the first the describe the pulmonary circulation. As Haddad and Khairallah (1936) have pointed out, the first description of the minor circulation was given in the XIIIth century in a manuscript commentary on the anatomy of the Canon of Avicenna by Ibn Nafis an Arabian physician who was dean of the Mansoury Hospital in Cairo. The description by Ibn Nafis as translated by Haddad and Khairallah gives a clear account of the pulmonary circulation:—

This cavity (where the blood is thinned) is the right cavity of the two cavities of the heart. If the blood is thinned in this cavity it must of necessity pass to the left cavity where the spirit is created. But between these two cavities there is no passage as that part of the heart is closed and has no apparent openings as some believed and no non-apparent opening fit for the passage of this blood as Galen believed. The pores of the heart there are obliterated and its body thick, and there is no doubt that the blood, when thinned, passes in the vena arteriosa to the lung to permeate its substance and mingle with the air, its thinned part purified; and then passes in the arteria venosa to reach the left cavity of the two cavities of the heart; having mixed with the air and become fit for the creation of the spirit.

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THE RETRO-PERITONEAL DERMOID CYSTS

By Milroy Paul
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EVERY case of retro-peritoneal dermoid cyst is worthy of record, as the condition is one of great rarity and of considerable interest. The mode of origin of retro-peritoneal dermoid cysts has not been adequately explained and the problem may be clarified by the careful recording of a sufficient number of cases. The retro-peritoneal dermoid cyst can be distinguished from cysts of the kidneys, pancreas, spleen, ovary and the occasional abdominal testis by its being free from attachment to any of these organs. On the other hand, the retro-peritoneal dermoid cannot be clearly demarcated from the mesenteric dermoid since the mesenteric cyst (Higgins and Lloyd, 1924) assumes a retro-peritoneal position when it becomes larger, while the retro-peritoneal dermoid may grow forwards into the mesentery. The subject of mesenteric dermoid cysts has been well reviewed by James (1944).

In any discussion on the origin of these cysts, the problem of definition of a dermoid cyst arises. A dermoid cyst has been demarcated from an epidermoid cyst by the fact that the epidermoid cyst is lined by a simple layer of flattened squamous epithelium and may contain serous fluid, while a dermoid cyst is lined by squamous epithelium showing fully formed or rudimentary dermal papillae, sebaceous glands, hair and sweat follicles, and it contains sebaceous material. This distinction between epidermoid cysts and the dermoid cysts, however, cannot be maintained, for sebaceous material has been found in many cysts lined by a simple layer of flattened squamous epithelium. In the case recorded in this paper, the cyst was lined by a simple layer of flattened squamous epithelium and yet it contained thick sebaceous material; Williams (1941) records a similar case. These case reports show that the lining membrane of a cyst containing sebaceous material may be of any grade of complexity from fully formed skin to a simple flattened squamous epithelial lining. Many of the cysts have shown the structure typical of a teratomatous dermoid (Gordon, 1925). Teeth and hairs are seen on a thickened plaque of the cyst wall, and examination of the plaque has demonstrated various structures such as bone, cartilage, thyroid tissue and intestine. These complex dermoid cysts, whether they are tridermal or bidermal in origin, could only have originated as teratomas. As the ovary and the testis are not involved, the teratoma can be accounted for by the blastomere theory of Marchand and Bonnet (1899), which dates histogenesis of these tumours from the earliest segmentation of the fertilized ovum. This is a more reasonable explanation of the origin of such tumours than the possibility of an origin from genital cells which had strayed from the germinal ridge (Felix, 1912). Even a unidermal teratoma could show complexity of tissues, as the cyst could give rise to teeth hairs and even rudiments of parts of the eye and central nervous system, but such cysts must be of teratomatous origin. Unidermal cysts, however, usually show only skin structures and one possible mode of origin of such cysts would be the sequestration of the skin during the process of development. This theory is, however, difficult to reconcile with the deep situation of the cyst in the posterior abdominal wall.

Dowd (1900) was responsible for focussing attention on the view first put forward by Roth (1881) that retro-peritoneal and mesenteric cysts could arise from vestiges of the Wolffian and Müllerian ducts or from pronephric or mesonephric tubules. In the case of cysts lined by stratified epithelium it is not likely that such cysts could have arisen from pronephric or mesonephric tubules, but the Wolffian duct which gives rise to the ureteric buds which are lined by

transitional epithelium, could quite well originate a flattened squamous cell lining for a dermoid. The Müllerian duct would, however, be an unlikely origin in the case of a female patient with fully developed Fallopian tubes and uterus as was the case in the patient reported on in this paper.

FIG. I. Photograph of patient before operation showing distension of abdominal wall by the large cyst.

The diagnosis of a retro-peritoneal cyst must always be difficult. In the case recorded in this paper, the absence of movement of the cyst on respiration served to exclude the diagnosis of a kidney tumour, and the large round mass was correctly considered to be a retro-peritoneal, non-malignant tumour. Had an X-ray picture been taken, a dense shadow with a round edge might have been present and it may have been possible that an accurate guess would have been made that the cyst was a retro-peritoneal dermoid.

Enucleation of the cyst is the treatment of choice, and in the cases where this was possible, the method has given uniformly good results (Handfield Jones, 1924). Marsupialization and drainage, which was probably first employed by Pean (1880), has also given good results in many recorded cases.

CASE REPORT

P.M.M.N., aged 60, a female married Sinhalese, was admitted to the General Hospital, Colombo, on 16th June, 1947, for a large abdominal tumour which had been noted for the past fifteen years. The tumour had already increased in size and it had not been painful or tender at any time. On examination, the patient was seen to be a well preserved, active woman (Fig. I). She had a large, round tumour extending from the left loin to the anterior abdominal wall which was bulged forwards by the tumour (Fig. I). It extended well beyond the middle line and both recti muscles were clearly seen on its surface when the patient tensed her abdominal wall (Fig. II). It was roughly spherical in shape, being well defined and easily palpable. It lay just below the costal margin, and extended to about three fingers breadth above the groin. It was soft, fluctuant, and dull on percussion. It did not move on respiration. The absence of an edge ruled out the possibility of the tumour being a ptosed spleen, and the position of the cyst with its upper pole below the costal margin, and with the absence of movements with respiration excluded the diagnosis as an enlarged kidney. The tumour was diagnosed as a soft retro-peritoneal non-malignant tumour probably a lipoma, and an operation was advised.

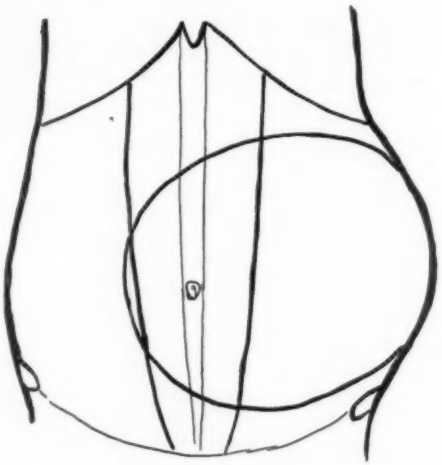

FIG. II. Diagram outlining area occupied by the cyst, and showing the recti muscles which were readily palpable in this case.

Operation under light percaine spinal anaesthesia on 26th June. Von Lichtenberg's incision (Robinson 1947), was employed extending forwards along the line of the twelfth rib. The twelfth rib was four inches long in this case, and it was resected sub-periosteally, the incision being continued on as far as the anterior axillary line. The abdominal muscles were divided in the line of the incision, and a retro-peritoneal encapsulated yellow tumour was seen. It was believed at this stage to

be a lipoma and enucleation of the tumour was now commenced by inserting the finger between the capsule of the tumour and its bed. The capsule tore while this was being done and thick grey cheesy fluid welled out. It was feared that a cold abscess had been inadvertently opened. The fluid content was emptied and the cyst wall wiped dry. The cyst wall was now seen to be quite well defined and to have no connection with the vertebral column. It was clear that the tumour was a large dermoid cyst, and enucleation of the cyst wall was continued. Anteriorly the wall of the cyst was so intimately blended with the peritoneum, as to make its removal by enucleation impossible. The peritoneal cavity was opened at this place and the peritoneum was removed with the cyst wall up to the place where the descending colon lay on the cyst. The descending colon was dissected away from the cyst wall. Above, the cyst touched the colic surface of the spleen but was not adherent to it. Below, the cyst lay in the iliac fossa. The pelvic viscera were clearly viewed through the opened peritoneum, and the left Fallopian tube and the ovary were seen in their normal position on the left side of the true pelvis. The left kidney lay behind the cyst. The cyst wall had a thickened, uniformly white fibrous plaque two inches in diameter on its anterior wall lateral to the line of the colon. The lining wall of the cyst was smooth and contained no hairs. The entire cyst wall was enucleated. The abdominal wound was closed without drainage.

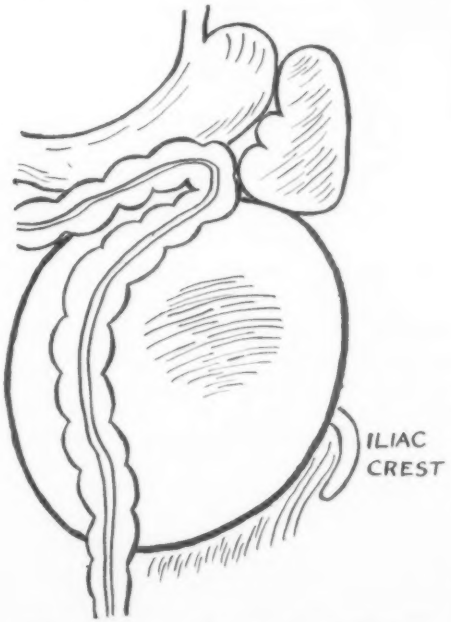

FIG. III. Diagram showing the principal relations of the cyst as viewed at operation. The thickened plaque on the anterior wall of the cyst is indicated by shading.

The patient had moderate abdominal distension for two days after operation but after that date she made an uninterrupted recovery leaving hospital on 9th July. She was seen again on 20th September and was in good health with no evidence of recurrence of the cyst.

Microscopic report: The cyst wall is lined by a layer of flattened squamous epithelium four or five cells deep. The basal cell layer shows a single row of dark staining round nuclei, while the other epidermal cells have light staining oval nuclei lying with their long axis in the line of the cyst wall. There is no evidence of cornification of the superficial layer of cells. The epithelial layer rests in a bed of hyalinized fibrous tissue in which there are a number of elongated connective tissue cell nuclei. There are no dermal papillae, sebaceous glands or sweat follicles (Fig. IV).

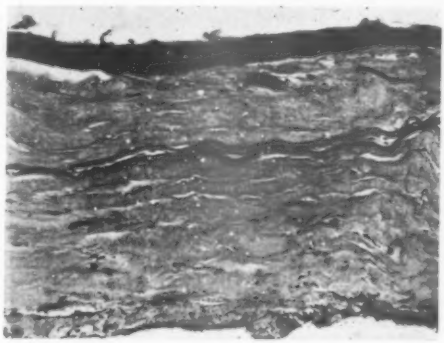

FIG. IV. Photomicrograph of cyst wall. The inner lining of squamous epithelium rests on a thick layer of fibrous tissue which has a single layer of endothelial cells on its peritoneal surface. (x 170)

COMMENTARY

The very large cyst described in this case was fully exposed through Von Lichtenberg's kidney incision, a result particularly noteworthy in that the twelfth rib was four inches long and would have hampered exposure had it not been removed. This incision was recommended by Robinson (1947) and it deserves to be more widely known.

The visualization of the left ovary and left Fallopian tube at operation in this case of left sided retro-peritoneal cyst, excluded the possibility of the cyst being an ovarian dermoid, and the absence of attachment except by areolar tissue to all structures

except the peritoneum on its anterior surface excludes the possibility of its having arisen from the kidneys, pancreas or spleen. The thickened plaque of fibrous tissue on the anterior wall of the cyst was underneath the area of firm attachment of the cyst wall to the peritoneum and was probably a chronic inflammatory thickening of the wall and not the embryonic disc of a teratomatous cyst.

The histological structure of the cyst wall is consistent with an origin from the Wolffian duct (Cave, 1925). A dermal origin is possible, and the simple epidermal structure of the wall is not against this mode of origin, for a similar structure is quite usual in cysts of undoubted dermoid origin, such as the dermoid cysts growing at the outer angle of the orbit.

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ARGENTAFFIN TUMOURS OF THE SMALL BOWEL AND VERMIFORM APPENDIX

By J. Freidin

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CARCINOID or argentaffin tumours form a well-defined but uncommon lesion which may affect any part of the gastro-intestinal tract but is more commonly encountered in the vermiform appendix and ileum.

It is important that this tumour be differentiated from the more frequently occurring carcinoma, not only by the pathologist but also by the surgeon, since the prognosis and treatment differ for the two conditions occurring at these sites.

A review of pathological material obtained from three Melbourne Hospitals is presented in order to emphasize the essential differentiating points of argentaffin tumours and the difficulty met in diagnosing them pre-operatively and in the post-mortem room.

Historical Note

Although Lubarsch (1888) first reported an unusual type of multiple tumour of the ileum occurring in the submucosa and histologically showing no glandular differentiation, it was not until Orberndorfer defined the tumour in 1907 and coined the term "carcinoid" that its essential features came to be realized.

In 1914 Masson and Gosset demonstrated the argyrophil nature of the granules in the cytoplasm of the cells forming these tumours and pointed to the similar characteristics exhibited by certain cells lining the glands of Lieberkühn; these were known as the yellow cells of Kultschitzky and were renamed "argentaffin" cells by Masson.

It is now generally accepted that carcinoids are argentaffin cell tumours.

The name carcinoid implied the occurrence of a tumour with definite histological features of local invasion but benign behaviour so far as metastases were concerned.

Since the report of definite metastasis of carcinoid tumours by Stewart and Taylor in 1926, a large number of cases has been presented, so that these tumours now merit the name "argentaffin-cell carcinoma."

Material

The material for this study was obtained from the records of the pathology departments of the Royal Melbourne, Alfred and Prince Henry's Hospitals in Melbourne.

The specimens cover a period of seventeen years from 1935 to 1952.

The number available totals 30 cases; of these 6 cases had been previously reported in 1940 by R. A. Willis. Of the 30 cases, 17 specimens were obtained at operation and 13 were obtained from post-mortem examination.

The material available for study consisted of histological sections stained with haematoxylin and eosin. A record of the macroscopic appearance was available in each case.

Analysis of Material

(a) Anatomical sites

There were 14 appendical argentaffin tumours, all removed by appendicectomy.

There were 16 cases where the primary lesion was extra-appendicular: of these three, all in the ileum, were found at operation; of the remaining 13, all obtained at post-mortem, two were in the duodenum, one in the jejunum and 10 in the ileum.

(b) Age incidence

In the appendicular argentaffin tumours the age varied from nine to 75 years with an average of 26.7 years.

In the extra-appendicular group the age varied from 37 to 77 years with an average of 61.5 years.

TABLE 1

Cases of extra-appendicular argentaffin tumours, with reference to the pre-operative diagnosis and the finding of primary and secondary growths.

Case	Pre-Operative Diagnosis	Primary Tumour	Secondary Tumour
1	Lump in right iliac fossa Carcinoma caeci	Ileum (single)	Mesentery
2	Acute appendicitis	Ileum (single)	Mesentery
3	Acute appendicitis	Ileum (single)	None

(c) Sex incidence

In the appendicular group there were 10 females and 4 males.

In the extra-appendicular group there were 2 females and 14 males.

(d) Method of presentation

None was diagnosed pre-operatively or, in the post-mortem cases, before death.

Of the 14 cases obtained by appendicectomy, 13 were resected during an emergency operation for acute appendicitis and one was removed during the course of an elective operation for retroversion of the uterus.

Of the 16 extra-appendicular cases three cases were operation specimens as shown in Table 1.

Of the remaining 13 extra-appendicular cases all of which were obtained at post-mortem the death of the patients could be attributed to metastasizing argentaffin-cell carcinoma in five instances, whereas in 8 cases the finding of argentaffin-cell carcinoma was supplementary to the lesion responsible for the patient's death.

These particulars are shown in Tables 2 and 3:

TABLE 2

The relation of single and multiple argentaffin tumours to the development of secondary growths in five post-mortem specimens considered to have caused the death of the patient.

Case (Autopsy)	Accompanying Lesion	Primary Tumour	Secondary Tumour
4	None	Ileum (multiple)	Liver, pancreas, spleen
5	Carcinoma of kidney	Ileum (single) (with peritonitis)	None
6	None	Duodenum (single)	Infiltrating pancreas
7	None	Ileum (multiple)	Mesentery, lung, pancreas, liver, kidney
8	None	Ileum (multiple)	Mesentery, pancreas, brain

TABLE 3

The relation of single and multiple argentaffin tumours to the development of metastases in eight post-mortem specimens considered to be incidental to the lesion responsible for the death of the patient.

Case (Autopsy)	Lesion Causing Death	Primary Argentaffin Tumour	Secondary Argentaffin Tumour
9	Carcinoma of bronchus with secondaries	Ileum (single)	None
10	Duodenal ulcer with haemorrhage	Ileum (multiple)	None
11	Chronic nephritis. Cerebral haemorrhage	Ileum (single)	Mesentery, liver
12	Myocardial infarction after resection for carcinoma of colon	Ileum (multiple)	None
13	Renal tract infection after prostatectomy	Ileum (multiple)	None
14	Secondary deposits from carcinoma of breast three years after radical mastectomy	Ileum (multiple)	None
15	Bronchopneumonia, renal tract infection after gastrectomy for benign ulcer	Duodenum (single)	Related node only
16	Metastasizing carcinoma of bronchus	Ileum (single)	None

(e) Macroscopic characteristics

The macroscopic appearances in this series follows the usual pattern that has been described for these tumours (Masson, 1928). The important features which help to differentiate them from adenocarcinoma are as follows:

(i) In the appendix the tumours are single, usually near the tip; the growth appears as a nodule in the wall leaving a normal lumen, or it may occur right round the wall causing much thickening with disappearance of the lumen. The tumour is hard, and yellow in colour.

In one case only in the present series did the tumour, situated about 1″ from the tip, produce obstruction with ballooning of the distal end of the appendix.

(ii) In the small intestine the tumours are often multiple and are to be regarded as multiple primary growths.

TABLE 4

The frequency of occurrence of single and multiple primary tumours in the operation and autopsy groups of extra-appendicular argentaffin tumours.

Source of Case	No. of Cases	Single Primary Tumour	Multiple Primary Tumour
Operation	3	3	—
Autopsy	13	6	7

(iii) The tumours appear macroscopically as "buttons" of white or yellowish firm tissue, occurring in the submucosa. This appearance results because the main bulk of the tumour is in the submucosa and the overlying mucous membrane does not often ulcerate.

This appearance is retained even when the tumour infiltrates more deeply through the muscle coats and appears on the serosal aspect. The lack of appreciation of the deep invasion on macroscopic appearance can be correlated with the microscopic finding that the columns of argentaffin cells infiltrate between and separate muscle bundles, rather than replace them in such a way that the architecture is destroyed.

FIG. I. Photograph of a specimen showing an argentaffin tumour of terminal ileum involving the ileo-caecal valve and projecting into the caecum, with large metastatic deposits (which were yellow in colour) in mesenteric lymph nodes. Operation specimen. (Case 1, Table 1)

It is of interest to note that this benign macroscopic appearance is the usual pattern seen even when metastases in mesentery, liver or pancreas are already demonstrable.

Another point that emerges from this study is the frequency with which the true nature of the ileal primary was not recognized: of 13 post-mortem cases the argentaffin nature of ileal "buttons" of growth was not recognized in seven cases till histological sections were examined; the diagnosis was made more difficult in some cases by the finding of another metastasizing neoplasm in the body so that the ileal growths were considered at first to be secondary tumours.

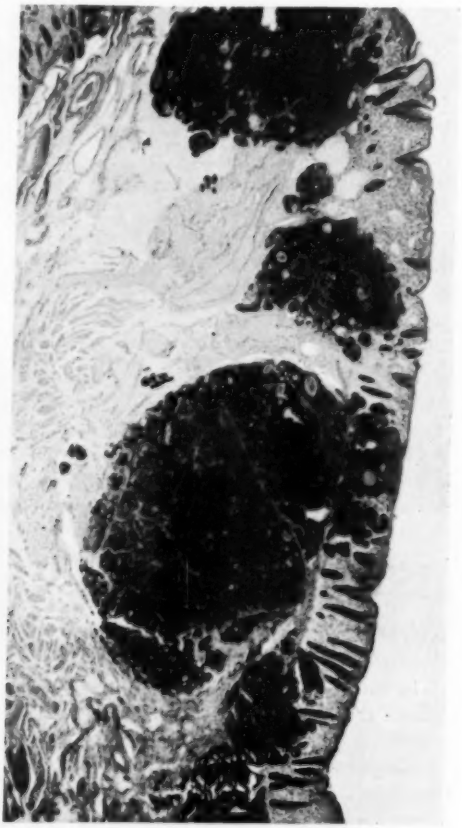

FIG. II. Photomicrograph of a section of the tumour shown in Fig. I. Columns of argentaffin cells collected in nodules. Note the intact overlying muscosa. (Case 1, Table 1) (x 20)

It is important, therefore, to appreciate the appearances of secondary sowing in the small bowel. When these occur as plaques of hard white tissue on the serosal aspect of the bowel, often with related plaques in the mesentery, little difficulty in diagnosis occurs. However, secondary

growths usually occur in the submucous coat either in sessile or pedunculated form. In the former case they are usually ulcerated, in contradistinction to the carcinoid tumours which usually are not. The pedunculated growths, as suggested above, present difficulties and, though the carcinoid tumours are sometimes yellow in colour, the problem is often only resolved by histological examination.

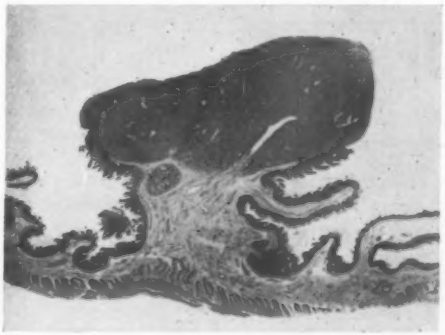

FIG. III. Photomicrograph showing a pedunculated argentaffin tumour of the ileum. Some surface ulceration is evident. Neoplastic cells have not infiltrated beyond the submucosa but metasases had occurred. (Case 4, Table 2) (x 5)

(f) Histological appearances

Argentaffin tumours present a characteristic pattern which can be recognized on routine staining with haematoxylin and eosin.

The cells are regular, round or polyhedral with a small round or oval nucleus: the nucleus is darkly staining, with a finely reticulated chromatin network; mitotic figures are very rare.

These cells are arranged in groups, irregular in outline but obviously forming the cross section of a column of these cells. Although in some instances a rosette formation may be distinguished, the important feature is that there is no evidence of glandular pattern and the mucin-producing cells of an adenocarcinoma are never seen.

The columns of argentaffin cells appear first in the mucosa between the mucous membrane and the *muscularis mucosae*. The growth is predominantly in this layer and as the nodule increases in size the *muscularis* becomes stretched and forms a pseudo-capsule; the mucous membrane continues intact over the surface of the nodule but may show some small areas of denudation. The type of neoplastic ulceration with leucocytic infiltration is not seen. Without any further change in the surface aspect of the growth the columns of cells invade through the *muscularis* into the submucosa and finally through the main muscle coats to the serosal aspect. In this process the columns appear to insinuate themselves between and thus separate the muscle bundles; thus the whole of the invaded wall is thickened; destruction of normal tissue is more commonly seen in secondary than in primary growths.

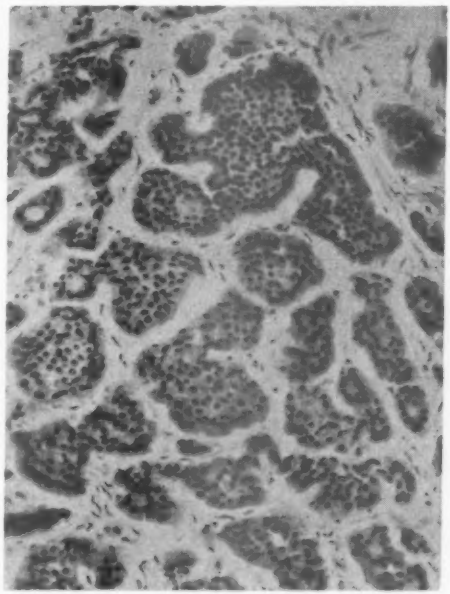

FIG. IV. Photomicrograph showing the typical arrangement of argentaffin cells in columns. The nuclei are round or oval, regular, without mitoses. Note the absence of glandular formation. (Case 4, Table 2) (x 150)

In no case is there any true capsule formation to be seen. The appendicular cases are in no way different from the small bowel cases and it is impossible to say from

histological examination which tumour would metastasize and which would not: histologically all are locally invasive. Nor is invasion through to the serosal layer essential for metastases as shown in Table 5.

In appendicular cases the average age is 26.7 years and the condition is more common in females, whereas in small bowel cases, the average age is 61.5 years and males are predominantly affected.

TABLE 5

The relation of depth of invasion to occurrence of metastases in the appendicular and extra-appendicular groups.

Anatomical Site	Involvement to Muscle Coat	Involvement to Serous Coat	Metastases
Appendicular	—	14	None
Extra-appendicular	5	—	3
	—	11	5

DISCUSSION

No case of argentaffin tumour was recorded in this series from any site in the gastro-intestinal tract apart from small bowel and appendix. Of the 16 small bowel cases only one was in the duodenum. This is in accordance with the usual frequency of these tumours at these sites, as reported in the literature (Grimes and Bell, 1949).

A definite change of opinion has occurred with regard to the benign nature of these tumours, since Forbus in 1925 classed them as essentially benign growths. This review further emphasizes the malignant behaviour of these tumours; although here only ileal tumours have shown metastases, metastasizing appendicular tumours have been reported (King, 1930). If an ileal tumour is found at operation, the wide resection appears essential for attempt at cure. It is reported in the literature (Grimes and Bell, 1949) that some beneficial effect can occur in these tumours when the primary is resected, even in the presence of metastases which cannot be removed.

From what "follow-up" was available for appendicular tumours further symptoms have not occurred and it is the general opinion that appendicectomy is sufficient in these cases.

There is also a noticeable difference between the appendicular and small bowel cases with respect to age and sex.

These differences in age, sex and metastasizing behaviour between appendicular and small bowel cases are difficult to correlate with their identical histological pattern.

SUMMARY

Thirty cases of argentaffin-cell carcinoma are reviewed.

They fall naturally into two groups depending on their anatomical site—the vermiform appendix or small bowel.

The incidence of age and sex in the two groups shows significant differences: young females are affected in the appendicular group, whereas elderly males are predominantly affected in the small bowel group.

It is noted that although the local invasive pattern in the bowel wall is identical, there is a gross difference in the frequency of metastases in the two groups.

The difficulties met in diagnosis before operation and in autopsy cases are exemplified.

The characteristic macroscopic and histological features of argentaffin tumours are described.

The surgical significance of these tumours is that the degree of malignancy in the different anatomical groups is very different and this influences treatment and prognosis.

ACKNOWLEDGEMENTS

I wish to thank the medical staffs of the Royal Melbourne, Alfred and Prince Henry's Hospitals for making the pathological material available. I am indebted to Mr. R. Inglis, photographer at the Royal Melbourne Hospital, for the photograph of Fig. I.

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Books Reviewed.

LOGAN TURNER'S DISEASES OF THE NOSE, THROAT AND EAR.

By DOUGLAS GUTHRIE and J. P. STEWART. Fifth Edition. Bristol: John Wright and Sons Ltd., 1952. 9" x 5½", xvi plus 468 pp., 246 illustrations, nine coloured plates. Price: 42s.

This book, appearing sixteen years after the previous edition, will be widely welcomed.

The editors have been assisted by six other well known oto-laryngologists of Edinburgh. The book is designed mainly for senior students and general practitioners. Although of moderate size it contains a very large amount of information due to a refreshing economy of language combined, for the most part, with clarity of style.

Excellent new articles include those on nasal physiology, injuries to the sinuses, aero-sinusitis and aero-otitis, pharyngitis in blood disorders, tests of cochlear and vestibular functions, teaching the deaf, hearing aids, and disorders of the voice.

The illustrations and coloured plates are very good.

Many oto-laryngologists, however, will not agree with some of the views expressed about prognosis and treatment. For example, concerning malignant tumours of the naso-pharynx the following statement appears. "The prognosis is good if the disease is recognised and treated at an early stage but—the disease is frequently overlooked until it is too late for radical treatment." We would never describe the prognosis of a malignant tumour of this region as good.

Another instance of unusual optimism is the following, appearing in the section on peroral endoscopy. "The aspiration and lavage of the bronchiectatic cavity or lung abscess give in many cases a complete cure of the disease."

An editorial oversight is responsible for confusion in the treatment recommended for acute suppuration of the frontal sinuses. After conservative treatment and two methods of draining the sinuses by external operation have been described, the following statement appears (a survival of the previous edition), "If these procedures should fail to relieve acute symptoms or if signs of orbital or intracranial inflammation supervene an external operation on the frontal sinuses will be necessary." The reader is then referred to page 90 which describes another method of surgical attack, which we would regard as contra-indicated in this condition.

Two other instances may be quoted of passages which have undeservedly survived from the previous edition. The first one appears under the heading Cerebrospinal Rhinorrhoea, the only reference to etiology being, "The exact cause is as yet uncertain . . ." The second instance, appearing on page 63, refers to the relationship of sinusitis to allergy, viz.: "In this connexion it must be mentioned that most observers agree that it is not the non-suppurative but the purely hyperplastic type of [sinus] infection that is associated with asthma and other allergic conditions." We have always found the meaning of this passage obscure, and the new spelling of "connexion" has not helped to clarify it.

Mention of Ferris Smith's external operation on the sinuses perpetuates that author's original error in the terminology of the ethmoidal vessels. He defines and ties the anterior, not the posterior, ethmoidal vessels.

Concerning tonsillectomy, we consider that undue prominence has been given to the "guillotine" method. Our own feeling is that this method of removing tonsils should be discontinued.

The treatment of laryngeal tuberculosis is dealt with at length but streptomycin is not mentioned in this section. It's use is, however, described in the section on antibiotics.

The operations described for the correction of outstanding ears do not include cartilage resection and would not give more than temporary improvement.

We feel that surgical approach to early eighth nerve tumours by the trans-labyrinthine route should not be mentioned, even to be partially condemned.

The unilateral nature of Meniere's disease is overstressed. In our experience a considerable proportion of these cases sooner or later develop bilateral involvement of the labyrinths.

We entirely disagree with the dictum in the section on antibiotics and sulphonamides, that when sulphonamides are being given in the treatment of otitis media, "It is advisable to stop all treatment after 4 or 5 days in order to observe the patient carefully for mastoiditis or other complication."

The doses of penicillin recommended for "severe" haemolytic streptococcal infections, namely 30,000 units in a watery solution three hourly, might in many cases be too small. Very much larger doses of combined crystalline and procaine penicillin given less frequently, can be more effective and are less disturbing to the patient.

BIOLOGIE, PATHOLIGIE UND THERAPIE DER GELENKE DARGESTELLT AM KNIEGELENK.

By ARNOLD SONNENSCHEIN. Basel, Switzerland: Benno Schwabe and Co., 1952. 9¾" x 7", xi plus 496 pp., 219 illustrations.

This book is a monumental monograph, devoted entirely to the knee joint. In a foreword the author explains the wider title it carries by stating that the knee joint can be taken as an example of human joints generally. In point of fact, as regards its anatomy and its mechanics, it cannot, and to that extent the title is misleading. Within the limitations stated it is a comprehensive and painstaking work of nearly 500 pages of closely written text.

The first 86 pages are devoted to general discussion of the joint—its morphology, development, mechanics and so forth. The 75 pages which follow deal with methods of investigation and examination.

The balance, except for a concluding 17 pages of bibliography, are given to the description, diagnosis and treatment of all the various ills and injuries which the human knee can suffer. Nothing would appear to have been left out. Methods of investigation include microscopic examination of aspirated fluid, and the endoscopic examination of the joint, as well as complete haematological and metabolic examination of the patient. Discussion of pathological conditions includes within its range the histology and pathology of rheumatic affections, and the relation of these to Selye's "adaptation syndrome."

Discussions on therapy include most of the modern methods, including the use of cortisone and B.A.L. in rheumatic affections. Surgical procedures are well described with a minimum of words, operative techniques being assisted by some well-chosen and excellently reproduced illustrations. Although some of the techniques recommended are more popular on the Continent of Europe than in this country, there is little in them that would be unknown to Australian surgeons. The author conveys in his discussions a general impression of a healthy conservatism in treatment.

The book is a valuable addition to the reference library. There is little of relevance that the enquirer will fail to find in it, and much of interest to orthopaedists and others. Its production is a credit to its publishers.

MODERN TRENDS IN NEUROLOGY.

By ANTHONY FEILING. London: Butterworth and Company Limited, 1951. 10½" x 7", ix plus 717 pp., 202 illustrations.

This is a most valuable volume for anybody interested in neurology and neurosurgery. The table of contents indicates that both Great Britain and the United States have been surveyed for the most worthy contributors.

The book takes a very wide sweep of the field of neurology and there are excellent contributions on all fields of advancing or changing thought. It is, however, far more than another "Recent Advances" type of work, as each author deals exhaustively with his subject—each chapter is a skilful consolidation of modern knowledge. Each section has a very full bibliography.

Denny Brown and Jackson Putnam make an excellent chapter on "The Frontal Lobes and their Functions," including a discussion of the surgical aspects.

Chapters on intracranial tumours, aneurysms, intervertebral discs, epilepsy, brachial neuralgia and special diagnostic methods, are all authoritative and expressive of the most recent opinion.

The general arrangement of headings and subheadings, beautiful type and ample illustrations greatly enhance its value.

It constitutes a very valuable book of reference and at the same time a work of easy and entraning interest.

ARCHITECTURAL PRINCIPLES IN ARTHRODESIS.

By H. A. BRITTAIN, O.B.E., M.A., M.Ch., F.R.C.S. Second Edition. England and Scotland: E. and S. Livingstone Ltd., 1952. 196 pp., 257 illustrations (nine in colour). Price: 42s. net.

This is the second edition of this well known monograph published ten years ago. No change has been made in the principles underlying operation but the author has kept an open mind and has made many changes in surgical technique as his experience developed.

The text of the new edition has been almost entirely rewritten and there are over one hundred more illustrations than before. Some chapters such as that on spinal fusion have been greatly enlarged and there are new operations for carpo-metacarpal arthrodesis and fusion of the symphysis pubis. To make the text more balanced and complete the author has included a chapter on the Naughton Dunn stabilisation of the foot which he practises without any modification.

The operation for ischio-femoral arthrodesis has fulfilled the author's promise and has received world wide recognition in the last ten years. In this edition he is able to elaborate his technical details and to give the results of one hundred and fifty-four operations with over eighty per cent. successful fusions of the hip.

This result in itself places this monograph on a high plane. The production by Livingstone maintains their recent standards in works of this kind and the new illustrations and better arrangement make it a more attractive volume than before.

LUMBAR DISC LESIONS.

By J. R. ARMSTRONG, F.R.C.S. Edinburgh and London: E. and S. Livingstone Ltd., 1952. 228 pp., 21 plates (coloured), 56 figures. Price: 42s. net.

There is possibly no more fascinating study in the problem encountered by orthopaedic surgeons than that of lumbar disc lesions, and although it is a bare twenty years since Mixter and Barr presented their historic comments to the Annual Meeting of the New England Society in Boston, a voluminous literature on this subject has made its appearance. There has not before, however, appeared such a complete and comprehensive presentation of the subject as this monograph by J. R. Armstrong. As Osmond-Clarke observes in his foreword to the book, this present monograph "is the product of accurate and inquisitive clinical observation, a comprehensive survey of the literature and an experience of more than a thousand well documented operations." This book is delightfully written, with an easy style which makes the reading of it not only an instruction, but indeed a pleasure.

In a series of seventeen chapters the anatomy functions, pathology, clinical picture, differential diagnosis and treatment of lumbar disc lesions are clearly and comprehensively dealt with.

In accordance with the usual high standard of the publishers, this book is beautifully produced with splendid radiographic reproductions and coloured, plain and line drawings.

The chapters on the function and pathology of lumbar disc lesions are particularly good, with many drawings to bring out points which are frequently ill-understood by many of the profession.

The section entitled "The Clinical Picture in Lumbar Disc Lesions" brings out the author's wide experience in the management of low back disorders, with points on the method of examination of patients and an explanation of the raison d'etre for the various signs. Possibly the best section is that of differential diagnosis. This is very full and on a considerably wider basis than is usual for this subject.

Treatment is considered in detail, both conservative and operative. It is refreshing to read that "The basis of any rational and effective treatment of a lumbar disc lesion is the establishment of the more favourable possible conditions for natural healing. While it is sometimes necessary to expose and remove the abnormal nucleus to achieve such conditions this is only so in a comparatively small proportion of all patients and the vast majority recover with effective conservative treatment."

Indications for operation are clearly given and emphasized and in describing the details of operative technique help is afforded by a series of beautiful coloured plates drawn from actual lesions discovered at the time of operation.

The final chapter deals with the recorded results of operative treatment. "The author has collected from the literature published between 1937 and 1949 the recorded results of operations performed on some four thousand patients by thirty-eight different surgeons or groups of surgeons." These figures are analysed.

The author then states that he himself "has now operated on more than one thousand patients with lumbar disc lesions." He further states that "any detailed analysis of the results would be out of place here but it is true to say that in general in his experience there is no other group in which a major surgical procedure has afforded so much satisfaction to both patient and surgeon."

This book will indeed be a valuable addition to the library of any surgeon who is called upon to deal with these recalcitrant problems.

A SYNOPSIS OF NEUROLOGY.

By W. F. TISSINGTON TATLOW, M.D., M.R.C.P., J. A. ARDIS, M.B., Ch.B., and J. A. R. BICKFORD, M.R.C.S., L.R.C.P. Bristol, England: John Wright and Sons Ltd., 1952. 7½" x 5", x plus 513 pp., 14 tables, 84 figures. Price: 30s. net.

As is usual with a synopsis, the authors have attempted to compress the whole subject of neurology into 468 pages. This impossible feat has necessitated the omission of several advances in the subject both as regards aetiology of diseases and methods of treatment. Diagnosis is stressed as would be expected in a British text book, but treatment which, of recent years has advanced considerably, is not given the attention it deserves. One has the impression that the student of this book would complacently fold his hands after arriving at a diagnosis, and feel that his work was done.

As examples, the treatment of trigeminal neuralgia is dismissed in a line or two with the text of which no modern neurosurgeon would agree. Migraine is discussed in some detail, but new methods of treatment of this disease are not mentioned. The advice —"surgical removal of the (cervical) disc" for pain in the upper extremity is too blunt, and may lead readers to think that this operation is routine.

The book is useful more particularly for the anatomical diagrams and physiological data; all of which could be obtained by the practitioner from current text books. However, it is of value—like the ready reckoner—but it is doubtful if the well trained neurologist or neurosurgeon would find it of great help.

It can be recommended to those cramming for a higher degree—as a companion to prescribed text books. It could also be recommended as a house physician's vade mecum. But like the meat extract, it fails to satisfy the intellectually and neurologically hungry.

CHIROPODIAL ORTHOPAEDICS.

By FRANKLIN CHARLESWORTH, F.Ch.S. Edinburgh: E. and S. Livingstone Ltd., 1951. 8¼" x 5¾", viii plus 255 pp., 131 illustrations. Price: 25s.

This monograph of 255 pages has no less than 131 figures and illustrations. It is in the latter that the chief merit of the work is to be found. They depict with remarkable clarity various foot deformities and the many ingenious appliances devised for their cure or relief.

There is certainly much in this work of great interest, and of sufficient importance to warrant frequent reference. The chapter on "Shoe Therapy" is excellent and might be studied with profit by professional and lay readers alike.

The chief criticism of the work is directed against the suggested use of a great variety of casts and appliances in the treatment of many conditions most of which can be far more efficiently —and to the patient, in the end, less expensively— treated by standard operative procedures.

There is, however, no doubt that a work of this nature has long been needed, and, as Sir Harry Platt states in a short foreword, should be of the greatest assistance to orthopaedic surgeons, physiotherapists, and chiropodists.

SURGICAL GYNAECOLOGY.

By J. P. GREENHILL. Chicago, U.S.A.: The Year Book Publishers, Inc., 1952. 8¼" x 5¾", 350 pp., 101 illustrations. Price: $8.50.

As the author states, this handbook consists, except for a section on pre-operative preparation and post-operative care and complications, entirely of drawings of operations and descriptions of the technique of these operations.

The methods which are described in detail are those used by the author, and almost the whole range of standard gynaecological operations is covered with the exception of the sling operations for stress incontinence.

The illustrations are excellent and the book should prove a most useful practical aid to house surgeons.

The description of haemorrhoidectomy is unlikely to be approved of by proctologists.